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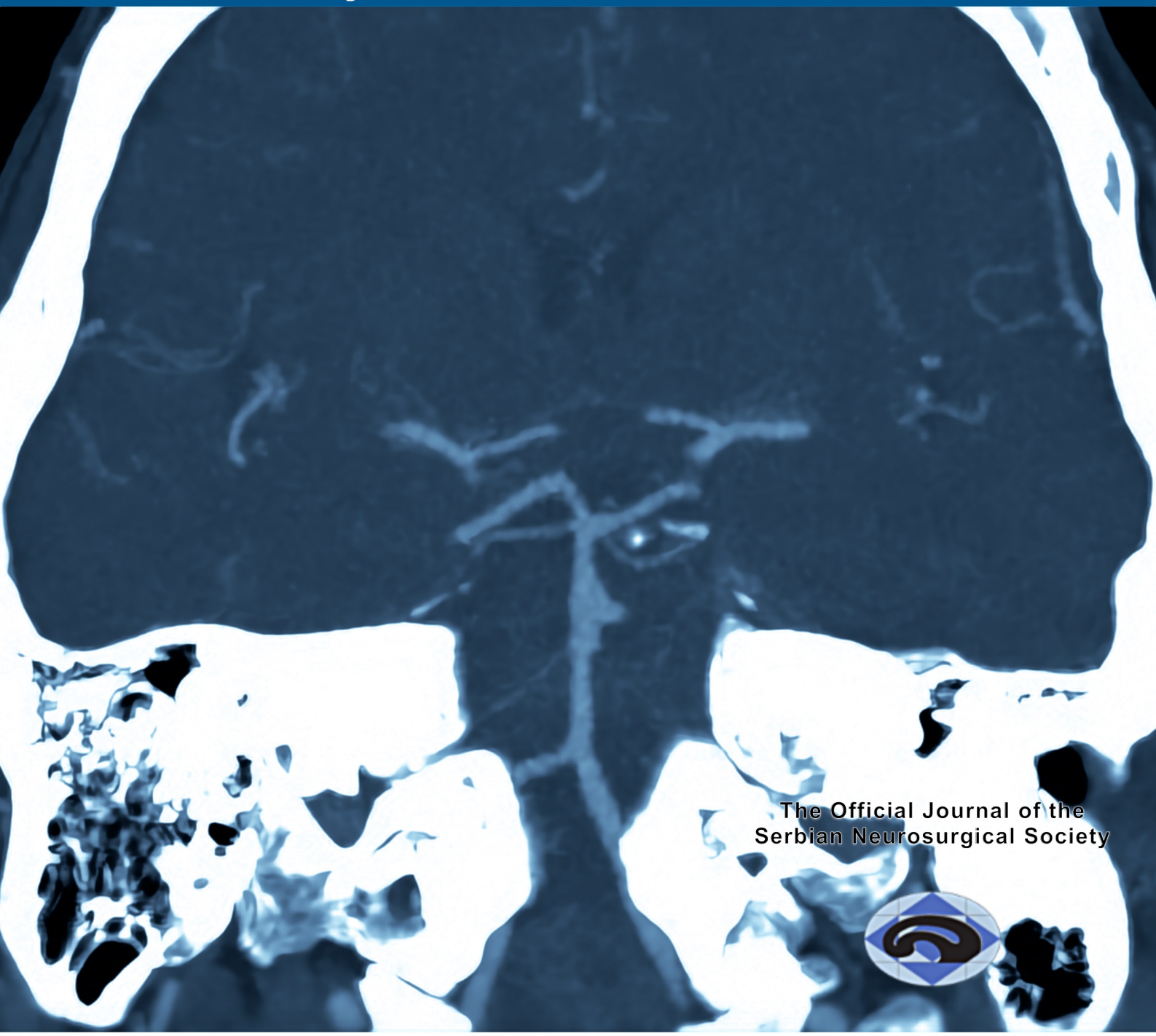

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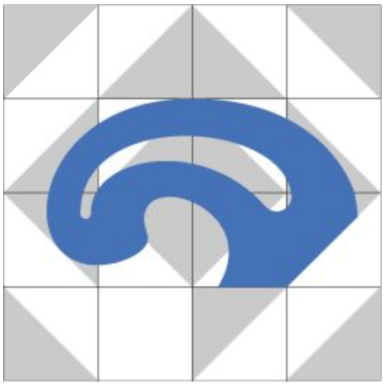

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Contents

Special Topic

NEUROSONOLOGY X NEUROSURGERY

EDITORIAL

The progress of "Our Journal" in anticipation of the centennial anniversary of neurosurgery in Serbia

Lukas Rasulić, MD, PhD[1], Milan Lepić, MD, PhD[1]

1. Neurohirurgija – The Serbian Journal of Neurosurgery

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This is the third year of *Neurohirurgija – The Serbian Journal of Neurosurgery*, which we usually informally refer to as "Our Journal". It was officially announced on October 31st, 2021, on the very date that marks the foundation of neurosurgery in Serbia. Our dedicated efforts towards establishing "Our Journal" had taken many years before its official launching, and we firmly believe that it was the right time to go ahead with it in anticipation of the centennial anniversary of neurosurgery in Serbia in 2023.

The first two years of "Our Journal" were marked by our struggle to attract papers, which we believed was the difficult part until we stumbled upon getting ignored by reviewers as our emails ended up in junk or spam folders, before ultimately having our website targeted by hackers and malware. We learned a lot through this experience and finally managed to achieve what could be considered the consolidated functioning of the Journal. We hope to keep up the pace and develop further into a reputable international journal in the field of neurosurgery and related borderline disciplines and subdisciplines.

So far, sixteen peer-reviewed papers were accepted and published (including this issue), by authors from Serbia and around the world, mainly from Europe. After the initial struggle, this year, we managed to publish most papers within a single issue, while significantly improving the time of accepting or declining decisions and acceptance/declining rates (***Table 1***).

Table 1. Editorial activity trends of Neurohirurgija – The Serbian Journal of Neurosurgery

	2023	2022	2021	Total
Submissions Received	4	14	5	23 (14/year)
Submissions Accepted	8	8	1	18 (8/year)
Submissions Declined	4	1	0	5
Submissions Declined (Desk Reject)	1	0	0	1 (0/year)
Submissions Declined (After Review)	3	1	0	4
Submissions Published	8	9	1	18 (9/year)
Days to First Editorial Decision	5	31	3	31
Days to Accept	32	165	88	160
Days to Reject	33	342	394	342
Acceptance Rate	75%	79%	80%	78%
Rejection Rate	25%	21%	20%	22%
Desk Reject Rate	0%	7%	0	4%
After Review Reject Rate	25%	14%	20%	17%

The latest issue encompasses three original research articles, one technical note within the Special Topic: "Neurosonology X Neurosurgery", one review of the literature with a series of cases and 3 reports of cases. Furthermore, all articles came from foreign authors, from Asia, Europe, and South America, with the special emphasize on two contributions from Ukraine and Russian Federation.

Our Journal has attracted attention from the readers' community as well, with many of the individual papers seen over 100 times, but also from other researchers, with 5 citations of 9 peer-reviewed articles published in the first two issues, which is already remarkable (***Table 2***).

Table 2. Abstract, full text views and citations of the papers published within the first two issues.

Article title	Abstract views	Full text views	Citations
Microsurgical management of complex middle cerebral artery aneurysms	314	124	1
Temporalis muscle reattachment by using transosseus running suture along superior temporal line: technical note	377	84	0
Motor nerve transfers for restoration of upper arm function in adult brachial plexus injuries	179	70	3
Why another journal? Well, why not? (Editorial)	130	66	-
Intraoperative ultrasound use in cranial neurosurgery	157	65	0
A state of art management of a bilateral basal ganglia germinoma: case report	110	54	0
Risk factors for postoperative infection after combat related head injuries	323	49	0
Delayed cerebral vasospasm following traumatic acute subdural hematoma: case report	119	40	0
Traumatic intracranial aneurysms associated with traffic accidents and endovascular management options	192	36	1
A warm Serbian welcome! (Editorial)	239	31	-
Dorsal hemangioblastoma with holocord syringomyelia: case report	88	29	0

There were two spikes in views, in August 2022 and March 2022, related to the announcements in the international mailing lists (***Figure 1***). Learning from this experience, we will persistently improve the spread in social media, with regular monthly updates.

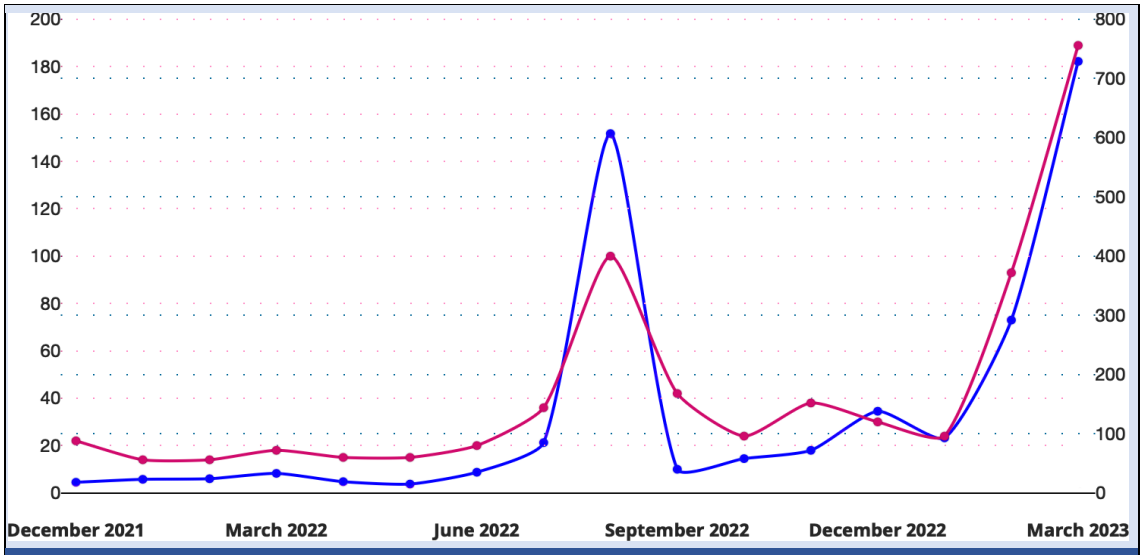

Figure 1. Monthly distribution of full text (pink) and abstract (blue) views since our first publication in December 2021.

In the forthcoming period, we will apply for the International Committee of Medical Journal Editors (ICMJE) listing as a journal that follows the ICMJE's Recommendations for the Conduct, Reporting, Editing, and Publication of Scholarly Work in Medical Journals, as well as for inclusion in the Directory of Open Access Journals (DOAJ), and once we reach the required number of published peer-reviewed articles, we will apply for indexing in PubMed Central and MEDLINE databases.

This October, Serbian neurosurgery will mark its Centennial Anniversary together with 85 years of the Clinic for Neurosurgery at the University Clinical Center of Serbia with the biggest neurosurgical gathering ever in Serbia, during the Neurosurgical Week from October 30th-November 3rd, 2023. The global theme of the meeting, *"Focus. Dedication. Specialization"*, sublimes the history of Serbian neurosurgery, as well as all our contemporary professional and academic activities and efforts in providing the best possible service and treatment standards to our patients and academic community.

We are looking forward to complementing this anniversary with the success of our very own Journal, *Neurohirurgija – The Serbian Journal of Neurosurgery*, as one of the landmark achievements of the Serbian Neurosurgical Society and Serbian neurosurgical community for the future generations.

ORIGINAL RESEARCH

The scapular notch: a Uruguayan cadaveric study of 62 dry scapulae

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Abstract

Introduction: The scapular notch is a depression on the superior border of the scapula, located medially to the coracoid process, through which suprascapular nerve enters the supraspinous fossa.

This paper aims to describe the main anatomical aspects of scapular notch, measuring anatomical parameters for identification of this region during surgical procedures, and compare the obtained data with previous worldwide publications.

Material and methods: Sixty-two dry scapulae of Uruguayan specimens were studied at the Anatomy Laboratory of the Faculty of Medicine, Universidad Centro Latinoamericano de Economía Humana (UCLAEH) in Maldonado, and the Faculty of Medicine, University of the Republic in Montevideo, Uruguay, and analyzed for variations.

Results: Of the 62 studied scapulae, 33 were right sided and 29 left sided. Anatomical variations were found in 19 specimens, which included 5 flattened shape notches (8.1%), and 14 ossified notches (22.6%), from which 4 (6.5%) were complete and 10 (16.1%) were incomplete. Scapular notch is located at an average distance of 66.7 mm (SD: 4.7) medially from the lateral border of the acromion.

Conclusions: Anatomy of the scapular notch is variable. The scapular notch can be located at the junction between the medial two thirds and the lateral one third of the superior scapular border. Anatomical variations of this region play an important role in the development of entrapment neuropathies and in surgical considerations for brachial plexus injuries reconstruction.

Keywords: scapular notch; suprascapular nerve; anatomy; variations; injury; entrapment

DOI: https://doi.org/10.55005/v3i1.2

Introduction

The suprascapular nerve (SSN) arises from the superior trunk of the brachial plexus. It runs posteriorly towards the superior border of the scapula and usually passes below the superior transverse scapular ligament (STSL) to reach the supraspinous fossa. In its further extent, the nerve turns downward to reach the infraspinous fossa, where it branches. The SSN provides motor innervation to the supraspinatus and infraspinatus muscles and also sends sensory branches to the coracohumeral and coracoacromial ligaments, subacromial bursae, and the acromioclavicular joint[1-4].

Compression of the SSN at the scapular notch (SN) is characterized by shoulder pain, and in severe cases, shoulder abduction and external rotation weakness in addition to supraspinatus and infraspinatus muscle atrophy[5]. Moreover, SSN is a common target in brachial plexus surgery for patients with impaired shoulder abduction, and the posterior approach for SSN neurotization is an alternative approach in some selected cases[6-8]. Therefore, a thorough knowledge of anatomical variations of SN, STSL, and the surrounding structures is imperative.

Materials and Methods

Sixty-two dry human adult cadaveric scapulae were investigated. The study was conducted at the Anatomy Laboratory of the Faculty of Medicine, Universidad Centro Latinoamericano de Economía Humana (UCLAEH) in Maldonado, (n=36) and the Faculty of Medicine, University of the Republic in Montevideo, (n=26), Uruguay.

The specimens were measured with a Vernier millimeter caliper (Mitutoyo Co., Tokyo, Japan). The following parameters were measured: A) Lower scapular angle to the lateral scapular angle, B) Lateral edge of the coracoid process to the superior scapular angle, C) Superior scapular angle to the lower angle, D) Depth of the SN, E) Width of the SN, and F) Lateral edge of acromion to the SN (***Figure 1***).

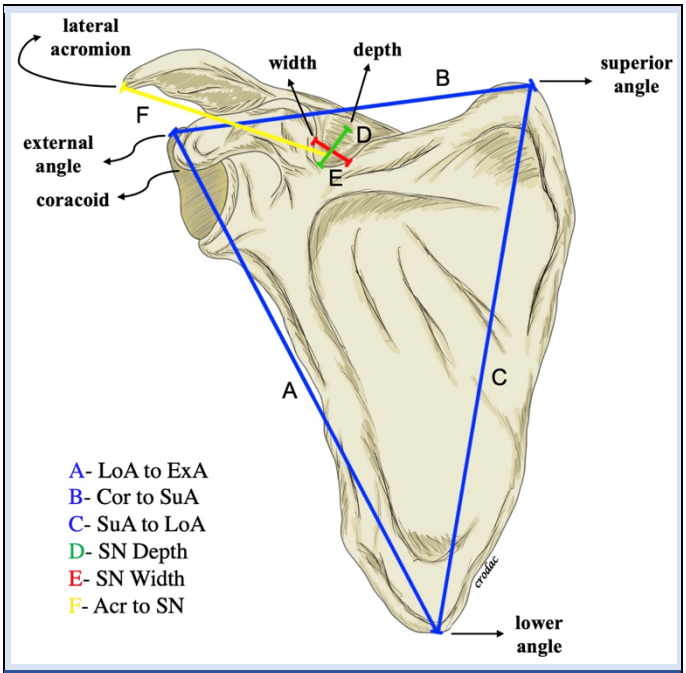

Figure 1. Scapular measured parameters: A) LoA to ExA, B) Cor to SuA, C) SuA to LoA, D) Depth of SN, E) Width of SN, F) Acr to SN. LoA, Lower angle; ExA, Lateral angle; Cor, Lateral coracoid edge; SuA, Superior angle; SN, Scapular notch; Acr, Lateral acromion edge

The average of all measurements and the standard deviation (SD) of width and depth of SN and the distance from the acromion to SN (F distance) were calculated. The relation between the length of the superior border of scapulae and F distance was analyzed. Statistical analysis of data was performed with the IBM SPSS Statistics v26 software.

Results

Of the 62 dry scapulae studied, 33 correspond to the right-side scapulae and 29 to the left-side scapulae. The obtained measurements (ranges, averages, and SD) are presented in ***Table 1***.

Table 1. Measurements of analyzed parameters.

Measure	Range	Average (SD)
lateral scapular border length	108-161	132.2 (9.9)
superior scapular border length	62-130	96.6 (8.1)
medial scapular border length	123-181	152.5 (15.8)
scapular notch depth	1-11	5.5 (2.2)
scapular notch width	2-14	7.5 (2.8)
lateral acromion to scapular notch distance	53-83	66.7 (4.7)

We recognized variations in 19 specimens (30.6%) that could make difficult the SN identification during surgical procedures or predispose to SSN entrapment at this level. The anatomical variations included SN flattening or osseous bridges (complete or incomplete). Of these morphological variants 8 (24.2%) were located on the right side, while 11 (37%) were on the left side. The difference between each side was not statistically significant.

We found a "nearly" flattened SN in 5 pieces (8.1%) and osseous bridges in 14 pieces (22.6%), of which 4 (6.5%) were complete (also known as scapular foramen) and 10 (16.1%) were incomplete.

A correlation between the length of the superior scapular border and distance from the acromion to SN was performed, and we identified that the distance from acromion to SN was 52 to 74% (average 61.3%) of the length of the superior scapular border. This means that the SN can be located at an average distance of 66.7 mm from the acromion (Table 1) or at the junction of the medial two-thirds and the lateral one-third of the superior border of the scapula.

Discussion

The SSN provides motor innervation to the supraspinatus and infraspinatus muscles and sensory innervation to the acromioclavicular joint. It reaches the supraspinous fossa passing through the SN, a bony landmark that is closed superiorly by the STSL, turning the SN into a fibro-osseous foramen[4,8-13] (***Figure 2***).

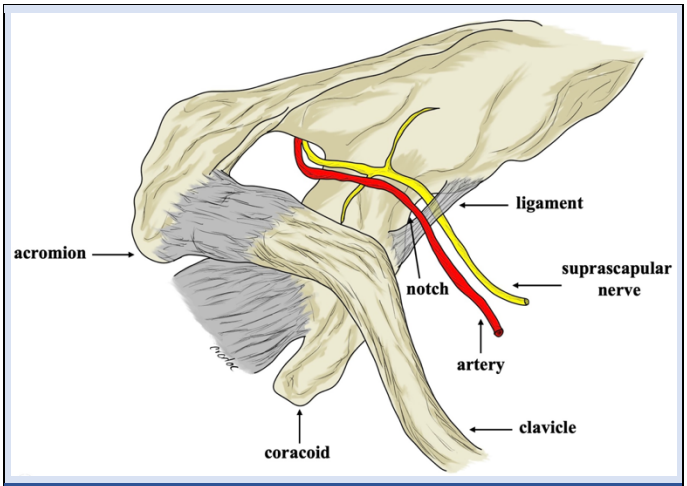

Figure 2. Scapular notch regional anatomy and content

This bony structure may be a potential site for entrapment or traction injuries in brachial plexus or isolated trauma of the SSN[2,4,15-17]. Many surgical procedures may require surgeons to approach the SSN distally to the SN. In consequence, anatomical variations of the SN, such as width, depth, anatomical orientation, osseous bridges, deserve anatomical and clinical considerations (***Figure 3A and 3B***).

Even though there are anatomical data that may seem irrelevant or redundant, each population has its own ethnic and anatomical variants. Before now, the anatomical variations of the scapular notch have not been studied in the Uruguayan population. Accordingly, it seems useful to contribute our findings to literature published from other countries[9,18,19].

The practical significance of the anatomy of the SN region lies in its application during brachial plexus reconstructive surgery, diagnosis and management of SSN neuropathy, anesthetic blockages of the nerve, and the risk of iatrogenic injury in some orthopedic surgical and non-surgical procedures[2,5-7,10,15,16,20,21].

In peripheral nerve surgery, the SSN is surgically treated for brachial plexus repair and its decompression for isolated entrapment neuropathy[6,22,23]. After dissection through trapezius and supraspinatus muscles fibers, the SN may be located by palpation[7,24]. However, the presence of an ossified STSL or a flattened scapular notch may render it difficult to localize this notch by palpation[22]. We observed such kind of anatomical variation in 14.5% of our specimens (8% flattened notch and 6.5% completely ossified STSL). The STSL should be divided to achieve nerve decompression, and facilitate its mobilization for repair in brachial plexus surgery. The presence of complete or incomplete ligament ossification adds greater difficulty to the surgical procedure[22].

The same consideration is applicable to SN entrapment neuropathy, for which it is necessary to divide the ligament for nerve release. A flattened notch or a completely ossified ligament increase the difficulty to identify the notch. Yavari et al. stated that 11.6% of 43 patients in whom a posterior surgical approach was performed for SSN neurotization had a "difficult" anatomy that made identifying the notch challenging[21]. They perform an osteotomy to release the SSN in 5 cases.

We found complete or partial ossification in 22.6% of our studied specimens, including 16.1% with an incomplete osseous bridge and 6.5% with a complete bridge. The incidence of complete ossification (scapular foramen) in our series is similar to French and Italian series[2,15,25]. A higher frequency of scapular foramen has been reported in in Indian (9.7-10%), European (5-6.1%) and North American populations (3.7-4%)[25]. The lowest prevalence of complete ossification of STSL is seen in Eskimo population (0.3%)[2,15], and the highest prevalence is seen in Brazilian population (30.7%)[22].

The prevalence of complete foramen in our study is similar to European population, which correlates with the ethnic distribution of Uruguayan population. In other words, 85.3% of our population is made up of Caucasian people, descendants of European immigrants, predominantly from Spain and Italy. Only 10.5% are dark-skinned and less than 5% are descendants of our aboriginal population (Charrúas, Chana-Timbúes, Guenoas, and Yaros)[26].

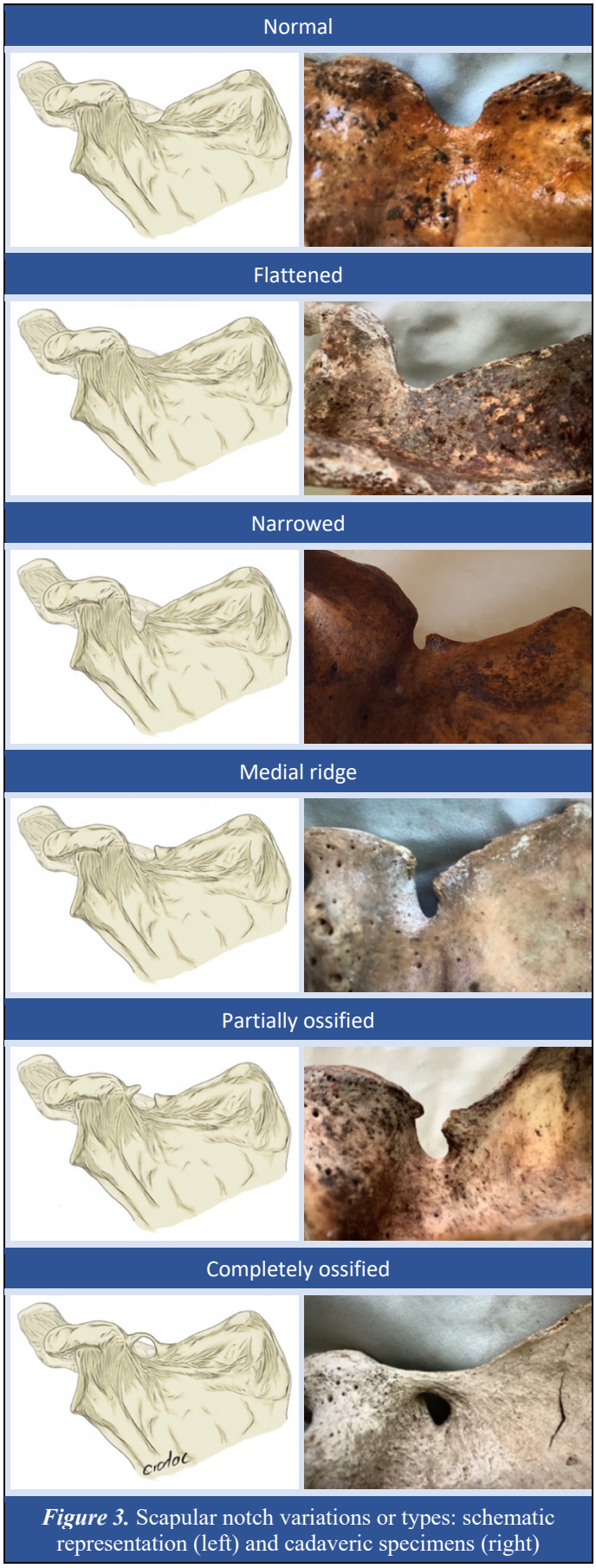

Figure 3. Scapular notch variations or types: schematic representation (left) and cadaveric specimens (right)

Tubbs et al. reported an incidence of 5-5.7% of ossified STSL, while other authors reported that 3.7-18% of human being have a partial ossification and 5-9.8% have a complete ossification[10,15,27-29]. It is paramount to recognize ossified ligaments during preoperative approach to properly remove them. Morphological features of the SN and SSN can be evaluated in real time with the aid of ultrasonography, especially in patients in whom surgical treatment has been considered[11](***Figure 4***).

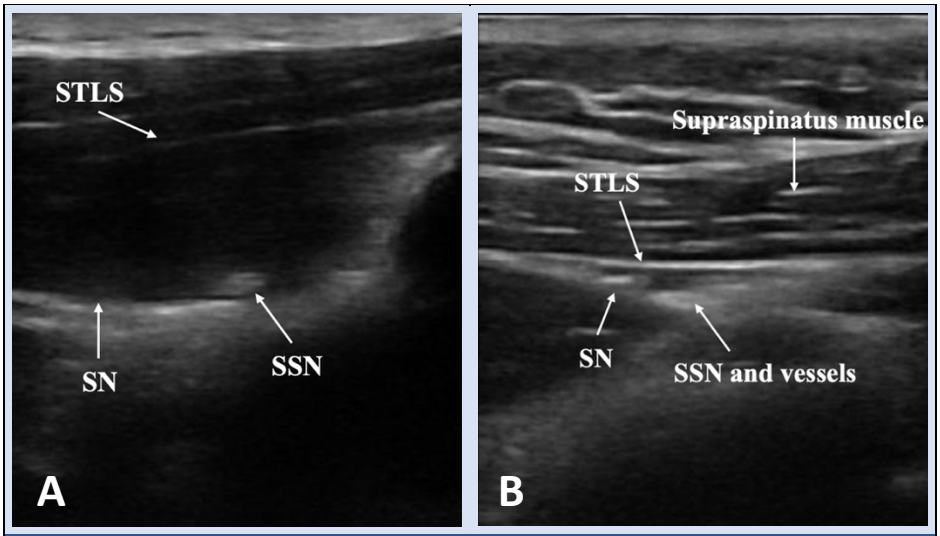

Figure 4. Ultrasonographic evaluation of scapular notch. A. Type II right sided normal SN with single unossified STLS. B. Type III left sided flattened SN with single unossified STLS. SN, Scapular notch; SSN, Suprascapular nerve; STLS, Superior transverse scapular ligament.

In a study conducted on dry scapulae and corpses, the authors carried out a histological study of cadaveric pieces with ossified ligaments and used corpses without an ossified ligament as control group. They found that in specimens with ossified ligaments, the SSN showed thickening of its diameter, fibrosis, and signs of nerve degeneration distal to the ligament. Therefore, they concluded that ossification of the STSL was a risk factor for the development of entrapment neuropathy[2]. However, a high incidence of ossified STSL does not proportionally correlate with the occurrence of SSN entrapment. Some researchers stated that SSN entrapment is responsible of painful shoulder in 0.3-2% of cases, and the incidence of ossification of STSL reaches 5-6% in general population. [2,4,5,9,10,13,20,27,29,32]. Ossification is a risk factor, but as expected, not the only responsible factor for nerve entrapment.

Regarding the "origin" of osseous bridges (osteophytes), Moriggl et al. suggest that its origin is related to endochondral ossification, as may occur with the Achilles tendon. Bony spurs are known to increase with age in the STSL, perhaps associated with the long-term effects of mechanical loading, as with Achilles tendon bony spurs or sesamoid bones in several body ligaments.

Moriggl et al. studied the cytoarchitecture of 7 STSL of adults with an average age of 57 years and ranged between 32 and 75 years, and described 3 cases with osteophytes, one of them with a sesamoid bone[16]. There is much evidence to support the idea that the formation of many bony spurs is triggered by the influence of mechanical forces over bones and ligaments. People with increased age, high weight bearing, or athletes are expected to have higher frequency of osseous bridges or complete foramina.

A second anatomical variation of STSL is the presence of a bifid or duplicated ligament. Polguj et al. described two anatomical forms of bifid STLS[15]. The first subtype has a superior and inferior band, and the second subtype has an anterior and posterior band (bifid in transverse plane). The incidence of double STSL reaches 3.1-3.8%, but the presence of an ossified double STSL is extremely rare. We did not find this anatomical variation in our sample, also considered a risk factor for SN entrapment[29].

Finally, data concerning gender dimorphism of the SN are controversial.

In a study of 812 specimens (cadaveric and radiological study of dry scapulae), Polguj et al. found a greater rate complete ossification of the ligament in men (6.4%) relative to 3.8% in women[9]. The same authors analyzed a dry scapulae sample in 2013 and observed complete ossification of the ligament in 7.4% of the cases. Females had a tendency for having the ligament ossified more frequently than men (9.1% vs 4.9% respectively). In a review, Polguj stated that males are more prone for having a calcified STSL, which may explain why SSN entrapment is more frequent in males. Nevertheless, more data is necessary to confirm if gender dimorphism increase the incidence of STSL ossification[21].

Another topic of interest is the content of the scapular foramen. Polguj et al. described four variations in the arrangement of the structures (SSN nerve, artery, and vein) passing through the SN[3]. In type I (61.3 %), the suprascapular artery travels above the STSL, while the suprascapular vein and SSN travel below. In type II (17 %) the two vessels traverse above the STSL, and the nerve is situated underneath the ligament. In type III (12.3 %), the suprascapular vessels and nerve lay underneath the ligament. For last, type IV (9.4 %) involves the combination of the other variants, albeit duplicated.

Specimens with a type III arrangement were found to have the smallest suprascapular opening diameter compared with the others. This may predispose patients to SSN entrapment, but further studies are needed to validate this finding[30]. SSN entrapment is complex and results from factors, such as the shape, length and width of the SN, the shape of the STSL (band or fan like), the presence of bifid STLS, anatomical variations of other ligamentous structures (anterior coracoscapular ligament), the number of neurovascular structures traversing the notch (such as in type III per the Polguj classification), the presence of hypertrophied suprascapular muscle, ossification degree of STSL, or the presence of tumors or "tumor like local pathology" (e.g., ganglion cysts)[3,9,12,31-33].

One interesting observation of our study is the F distance measurement from the lateral edge of the acromion to the center of the scapular notch. The average distance was 66.7 millimeters, matching with our previous anatomical and surgical studies, and is a key for the intraoperative localization of the SN. Previously, we postulated that in posterior surgical approach to SN decompression or neurotization with spinal accessory nerve, anatomical landmarks are of value[6]. We found that suprascapular and spinal accessory nerves are located at 70 and 110 millimeters, respectively, medially to the lateral aspect of the acromion. Our findings in this series confirm our previous data in a small sample of cadaveric specimens.

An incision to performing a spinal accessory nerve to SN neurotization should include these two anatomical landmarks. When we studied the correlation between the distance from acromion to SN/length of the superior border of the scapula, we found that distance from acromion to SN is 61% of the length of superior border of the scapula. The SN can be located at the junction of the medial two-thirds with the lateral one-third of the superior scapular border.

Conclusions

Anatomical variations in the morphology of SN may influence the development of SSN entrapment, but the information obtained by anatomical reports is contradictory. Ossification of STSL is frequent in general population (3.7 to 9.8%), whereas the incidence of SN entrapment is very low. Cadaveric anatomical studies indicate that fibrosis and nerve degeneration are proportional to the ossification degree of STSL, though it is not possible to link these findings to a clinically significant SN entrapment syndrome. Future studies examining brachial plexus surgery and SN decompression should consider these anatomical variations.

Disclosures

Conflict of Interest: All authors certify that they have no affiliations with or involvement in any organization or entity with any financial interest (such as honoraria; educational grants; participation in speakers' bureaus; membership, employment, consultancies, stock ownership, or other equity interest; and expert testimony or patent-licensing arrangements), or non-financial interest (such as personal or professional relationships, affiliations, knowledge or beliefs) in the subject matter or materials discussed in this manuscript.

Ethical approval: All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

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References

1. Basta M, Sanganeria T, Varacallo M. Anatomy, Shoulder and Upper Limb, Suprascapular Nerve. [Updated 2021 Oct 9]. In: StatPearls [Internet]. Treasure Island (FL): StatPearls Publishing; Acces 8/1/2022
2. Tubbs RS, Nechtman C, D´Antoni AV, et al. Ossification of the suprascapular ligament: A risk factor for suprascapular nerve compression? Int j Shoulder Surg 2013;7(1):19-22. doi: 10.4103/0973-6042.109882
3. Polguj M, Rozniecki J, Sibinski M, Grzegorzewski A, Majos A, Topol M. The variable morphology of suprascapular nerve and vessels at suprascapular notch: a proposal for classification and its potential clinical implications. Knee Surg Sports Traumatol Arthrosc 2015;23:1542–1548. doi.org/10.1007/s00167-014-2937-1
4. Das S, Suri R, Kapur V. Ossification of superior transverse scapular ligament and its clinical implications. Sultan Qaboos Univ Med J 2007;7(2):157–160.
5. Martínez F: Neuropatía del nervio supraescapular: reporte de dos casos. Rev Urug Med Int 2018; 2: 38-43. doi: 10.26445/rmu.3.2.5
6. Martínez F. Technical Note: Spinal-Accessory to suprascapular nerve transfer by posterior approach. Austin Neurosurg Open Access 2017;4(1):1058.
7. Martinez F, Jaume A, Sienra C, Russo A. Anatomía quirurgica de la transferencia nerviosa de espinal accesorio a nervio supraescapular por vía posterior. Neurocirugía/Neurocirurgia (FLANC) 2017;26(1):1-6.
8. Martinez Benia F. Transferencia nerviosa de accesorio-espinal a supraescapular: comparación de vía anterior versus vía posterior. Presentación de una serie y revisión de la literatura. Rev Arg Neurocir 2022;36(1):13-20.
9. Polguj MB, Sibinski M, Grzegorzewski A, Waszczykowski MB, Majos A, Topol MB. Morphological and Radiological Study of Ossified Superior Transverse Scapular Ligament as Potential Risk Factor of Suprascapular Nerve Entrapment. BioMed Res Int 2014; doi: 10.1155/2014/613601
10. Polguj MB, Sibinski M, Grzegorzewski A, Grzelak P, StefaNczyk L, Topol M. Suprascapular Notch Asymmetry: A Study on 311 Patients. BioMed Research Int 2014, Article ID 196896.
11. Jezierski H, Podgórski M, Wysiadecki G, Olewnik Ł, De Caro R, Macchi V, Polguj M. Morphological Aspects in Ultrasound Visualization of the Suprascapular Notch Region: A Study Based on a New Four-Step Protocol. J Clin Med 2018;7(12):491. doi: 10.3390/jcm7120491
12. Kumar A, Sharma A, Singh P. Anatomical study of the suprascapular notch: quantitative analysis and clinical considerations for suprascapular nerve entrapment. Singapore Med J 2014;55(1):41-4. doi: 10.11622/smedj.2014009
13. Bayramoğlu A, Demiryürek D, Tüccar E, Erbil M, Aldur MM, Tetik O, Doral MN. Variations in anatomy at the suprascapular notch possibly causing suprascapular nerve entrapment: an anatomical study. Knee Surg Sports Traumatol Arthrosc 2003;11(6):393-8. DOI: 10.1007/s00167-003-0378-3
14. Sangam MR, Sarada Devi SS, Krupadanam K, Anasuya K. A study on the morphology of the suprascapular notch and its distance from the glenoid cavity. J Clin Diagn Res 2013;7(2):189-92. doi: 10.7860/JCDR/2013/4838.2723
15. Polguj M, Jędrzejewski K, Majos A, Topol M. Variations in bifid superior transverse scapular ligament as a possible factor of suprascapular entrapment: an anatomical study. International Orthopaedics (SICOT) (2012) 36:2095–2100. doi.org/10.1007/s00264-012-1618-2
16. Moriggl B, Jax P, Milz S, Büttner A, Benjamin M. Fibrocartilage at the entheses of the suprascapular (superior transverse scapular) ligament of man—a ligament spanning two regions of a single bone. J Anat 2001;199(Pt 5):539-45. DOI: 10.1046/j.1469-7580.2001.19950539.x
17. Ohanisian L, Brown N, White SD, Rubay D, Schwartz PM. Persistent Shoulder Pain Due to a Suprascapular Nerve Injury in the Setting of Trauma. Cureus 2019;11(3):e4224. doi: 10.7759/cureus.4224
18. Daripelli S, Tolupunoori B, Vinodini L. Morphometric Study of Suprascapular Notch and Its Safe Zone in Indian Population. Maedica (Bucur) 2020;15(4):461-467. doi: 10.26574/maedica.2020.15.4.461
19. Long R, Wang N, Liu W, Liu Z, Cheng S, Zhang X, Geng X, Liu C, Wang J, Ding M, Tian Z, Li J. An anatomical study of the superior transverse scapular ligament of Jining population. Surg Radiol Anat 2019;41(11):1345-1349. DOI: 10.1007/s00276-019-02330-8
20. Sinkeet SR, Awori KO, Odula PO, Ogeng'o JA, Mwachaka PM. The suprascapular notch: its morphology and distance from the glenoid cavity in a Kenyan population. Folia Morphol (Warsz) 2010;69(4):241-5.
21. Polguj M, Jędrzejewski KS, Topol M. Sexual dimorphism of the suprascapular notch – morphometric study. Arch Med Sci 2013;9(1):177-83. DOI: 10.5114/aoms.2013.33173
22. Yavari M, Hassanpour SE, Alizadeh Otaghvar H, Abdolrazaghi HA, Farhoud AR. The Incidence of Ossified Superior Transverse Scapular Ligament during Nerve Transfer through Posterior Shoulder Approach. Arch Bone Jt Surg 2019;7(3):258-262.
23. Bhandari PS, Sadhotra LP, Bhargava P, Singh M, Mukherjee MK, Bhatoe HS. Dorsal approach in spinal accessory to suprascapular nerve transfer in brachial plexus injuries: technique details. Indian J Neurotrauma 2010;7(1):71-4. doi: 10.1016/S0973-0508%2810%2980015-2
24. Colbert SH, Mackinnon S. Posterior approach for double nerve transfer for restoration of shoulder function in upper brachial plexus palsy. Hand (N Y). 2006;1(2):71-7. doi: 10.1007/s11552-006-9004-4
25. Agrawal D, Singh B, Dixit SG, Ghatak S, Bharadwaj N, Gupta R, Agrawal GA, Nayyar AK. Morphometry and variations of the human suprascapular notch. Morphologie 2015;99(327):132-40. DOI: 10.1016/j.morpho.2015.04.002
26. Kluver C, Naranja Sotelo M. Ascendencia – Étnica y Seguridad Social en Uruguay. Asesoría en Políticas de Seguridad Social. 2019. Accessed: 12.6.2022: https://www.bps.gub.uy/bps/file/16400/1/65.-ascendencia-etnica-y-seguridad-social-en-uruguay.-c.-kluver-y-m.-naranja-sotelo.pdf
27. Kannan U, Kannan NS, Anbalagan J, Rao S. Morphometric study of suprascapular notch in Indian dry scapulae with specific reference to the incidence of completely ossified superior transverse scapular ligament. J Clin Diag Res 2014;8(3):7-10. doi: 10.7860/JCDR/2014/7836.4089
28. Albino P, Carbone S, Candela V, Arceri V, Vestri AR, Gumina S. Morphometry of the suprascapular notch: correlation with scapular dimensions and clinical relevance. BMC Musculoskelet Disord 2013; 14:172. doi.org/10.1186/1471-2474-14-172
29. Joy A, Sinha MB, Satapathy BC. The Ossified Bifid Superior Transverse Scapular Ligament Causing a Double Suprascapular Foramen: A Case Report. J Clin Diag Res 2015;9(6): AD03-AD04. doi: 10.7860/JCDR/2015/12812.6037
30. Yang HJ, Gil YC, Jin JD, Ahn SV, Lee HY. Topographical anatomy of the suprascapular nerve and vessels at the suprascapular notch. Clin Anat. 2012;25(3):359-65. DOI: 10.1002/ca.21248
31. Kharay SS, Sharma A, Singh P. Unusual morphology of scapulae: incidence and dimensions of ossified ligaments and supraspinous bony tunnels for clinical consideration. Singapore Med J 2016;57(1):29-32. doi: 10.11622/smedj.2015103
32. Bagoji IB, Hadimani GA, Bulgoud RS, Desai V, Prakash KG, Bharatha A. Anatomical Variations of the Suprascapular Notch and its Importance in Suprascapular Entrapment Neuropathy. Maedica (Bucur) 2020;15(3):298-304. doi: 10.26574/maedica.2020.15.3.298
33. Łabętowicz P, Synder M, Wojciechowski M, Orczyk K, Jezierski H, Topol M, Polguj M. Protective and Predisposing Morphological Factors in Suprascapular Nerve Entrapment Syndrome: A Fundamental Review Based on Recent Observations. Biomed Res Int 2017;:4659761. doi: 10.1155/2017/4659761

ORIGINAL RESEARCH

Neurosurgical service during COVID-19 pandemics in Ljubljana, Slovenia- lessons learned

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Abstract

Introduction: The novel coronavirus disease 2019 (COVID-19) became an important and urgent threat to global health. In Slovenia, the COVID-19 struck the health system immensely. Neurosurgery experienced difficulties, not only in regular, elective surgeries, but also during emergency situations.

Methods: In the article, we analyse and compare the number of elective and emergency neurosurgical procedures during the time of the pandemic (from March 2018 to February 2020) and describe our protocol in the management of neurosurgical patients in the Medical Centre in Ljubljana, Slovenia.

Results: There were 2597 patients treated surgically, including 1932 emergency patients and 665 emergency patients. Overall, we recorded an 11.2% drop in all neurosurgical procedures in two years after COVID-19 was declared compared to two years before. Elective procedures decreased by 13.9%, mostly on account of spinal pathology procedures (245, 23.5%), functional neurosurgical procedures (37 cases, 24.7%), endonasal endoscopy procedures (11, 12.8%), and brain lesions (31, 4.8%).

Conclusion: COVID-19 had a vast impact on the healthcare system in Slovenia, including on neurosurgery. New and improved strategies to maintain neurosurgical practice during public health emergencies are necessary for the neurosurgical service and healthcare system to run smoothly in the long term and prevent disruptions during future pandemics.

Keywords: COVID-19; neurosurgery; patient management; antivirus protocol; pandemic measures; Ljubljana

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Introduction

COVID-19 is a novel coronavirus disease caused by severe acute respiratory syndrome coronavirus-2 (SARS-CoV-2)[1,2]. Although it primarily affects the respiratory system, other organs, including the brain, may be affected. SARS-CoV-2 was first reported as four cases of pneumonia of unknown etiology on December 29th, 2019, in Wuhan, Hubei Province in China and is responsible for an ongoing pandemic. The disease presented with severe and unique biological characteristics, specific clinical symptoms, and particular blood test results and imaging features[2-4]. As a result of a rapid spread due to high transmissibility, the virus was recognized as a major threat to global health on March 11th 2020, when COVID-19 was declared a pandemic by World Health Organization.

At the time of writing in August 2022, the number of infected patients exceeded 550 million in almost every country around the world. The real number of infected people is in all likelihood much higher. The number of deaths caused by COVID-19 is predicted to be more than 6.4 million[1,5].

The COVID-19 pandemic caused difficulties in every health system. European countries were almost equally affected and adjustments were necessary to provide care for COVID-19 patients and normal functioning of the health system in parallel[6]. The European Union issued recommendations for member states in addressing pandemics. Of course, the European countries differed in their health policies.

Many departmental protocols were implemented, and protective measures were taken to cope with the massive influx of COVID-19 patients while preserving the regular medical services running normally[7,8]. Medical staff, equipment, and material were reallocated; management protocols were created; and dedicated in-hospital routes and operating theatres were established for ill patients[6,9]. Strict control of elective and/or emergency admissions, prevention of intermixing of cases and health care staff, improvements in operation and treatment processes, and strict ward management rules were put into practice[7,9,10]. The conventional outpatient service was altered into a telemedicine outpatient service, and elective surgeries were postponed or stopped. Moreover, some patients with COVID-19 infection had to undergo vital surgery, while others became symptomatic within days of elective surgeries[6,7,10,11].

Slovenia is a central European country with a population of approximately two million inhabitants. There are two neurosurgical centers, one in Ljubljana, the capital city, and the other in Maribor, the second largest city. Both are organized as departments within the Division of Surgery, located at university hospitals. The Department of Neurosurgery at University Medical Centre Ljubljana comprises 50 beds and an intensive care unit, while the one in Maribor is half the size. The Department of Neurosurgery in Ljubljana covers approximately two-thirds of the population, while Maribor covers the rest.

Compared to other European countries, COVID-19 struck our health system immensely. Due to rapid virus spread in Slovenia, it was practically impossible to make substantial timely preparations to minimize the impact of the pandemic and adapt the health system quickly. The official anti-virus measures at the state level were put into action a few days after the first case was confirmed, on March 4th, 2020, and the pandemic was declared a week after[12]. In addition, the general pandemic measures on the state level, including state lockdown, mandatory masks, hand disinfection, testing, and strict border crossing rules, a series of hospital and departmental protocols were instituted to limit the virus spread inside the hospitals and health centers. Some of the measures included adjustments of outpatient services, strict regulations upon patient admission, additional departmental and surgical premises for high-risk or COVID-19 positive patients, quantitative reduction of the surgical program including postponement of elective surgeries, reallocation of staff to COVID-19-related posts, constant use of protective equipment, and expansion of the telephone consultation service. Neurosurgery has also experienced difficulties, not only in regular, elective surgeries but also, especially during emergencies[12,13]. The management of these neurosurgical patients has therefore become more difficult than ever. In the article, we outline the impact that COVID-19 has had on the management of patients at the neurosurgical department in Ljubljana and describe our departmental protocols.

Methods and results

Patient analysis

We analysed and compared the number of elective and emergency surgeries before and during the pandemic. We denote an emergency case as any case that needed immediate surgery in a matter of hours due to a life-threatening situation and an elective case as any case that was scheduled in advance and operated on during working hours. Vital or emergency surgery encompassed any form of an acute brain haemorrhage (subdural, epidural or intracerebral), decompressive craniectomy, insertion of external ventricular drainage or intracranial pressure monitoring and evacuation of a chronic subdural haematoma or vital spinal pathology. For the analysis, we divided the procedures into subgroups: I) spinal pathology (spinal degenerative disorders, spinal lesions), II) brain lesions (any supra- or infratentorial brain tumours, cysts, abscesses or similar pathology), III) ventriculoperitoneal drainage, IV) cranioplasty, V) functional neurosurgical procedure (deep brain stimulation, spinal cord stimulation, vagus nerve stimulation, replacement of the batteries), VI) vascular pathology (aneurysms, arteriovenous malformations, and similar, VII) endonasal endoscopy (pituitary adenoma, Rathke cyst, clivus chordoma, craniopharyngioma).

We analysed the patient number and the pathology two years before the COVID-19 pandemic was declared (from March 2018 to February 2020). During this time, 2924 patients were treated surgically at the Department of neurosurgery in Ljubljana, including 2413 patients who represented elective cases. Overall, 1042 surgeries included spinal pathology, 646 brain lesions, 154 elective ventriculoperitoneal drainages for hydrocephalus treatment, 41 cranioplasties, 150 functional, 62 vascular, 86 endonasal endoscopic and 62 other procedures. There were 681 emergency cases.

During the two years of COVID-19 pandemic, from March 2020 to February 2022, there were 2597 patients treated surgically. Elective procedures included 1932 patients (***Figure 1***).

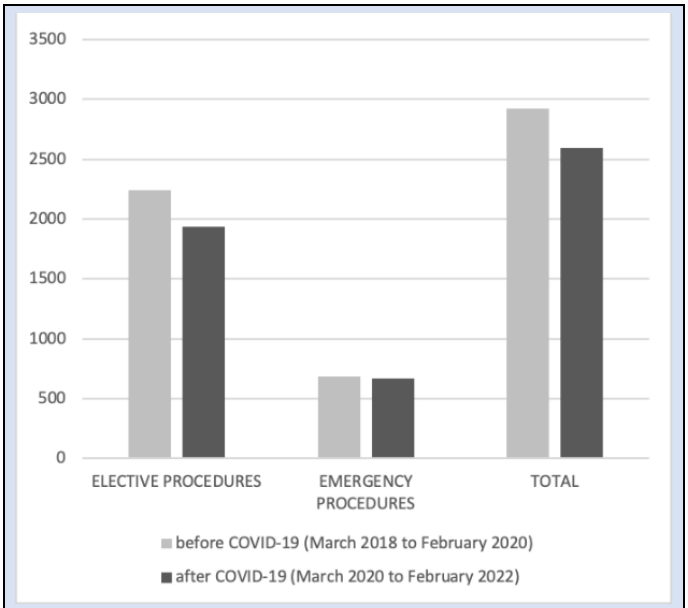

Figure 1. Elective and emergency neurosurgical procedures before and after the COVID-19 pandemic.

There were 797 spinal pathology surgeries, 665 brain lesions, 153 ventriculoperitoneal drainages, 50 cranioplasties, 113 functional, 65 vascular, 75 endoscopic endonasal and 64 other procedures. Emergencies were encountered in 665 cases (***Figure 2***).

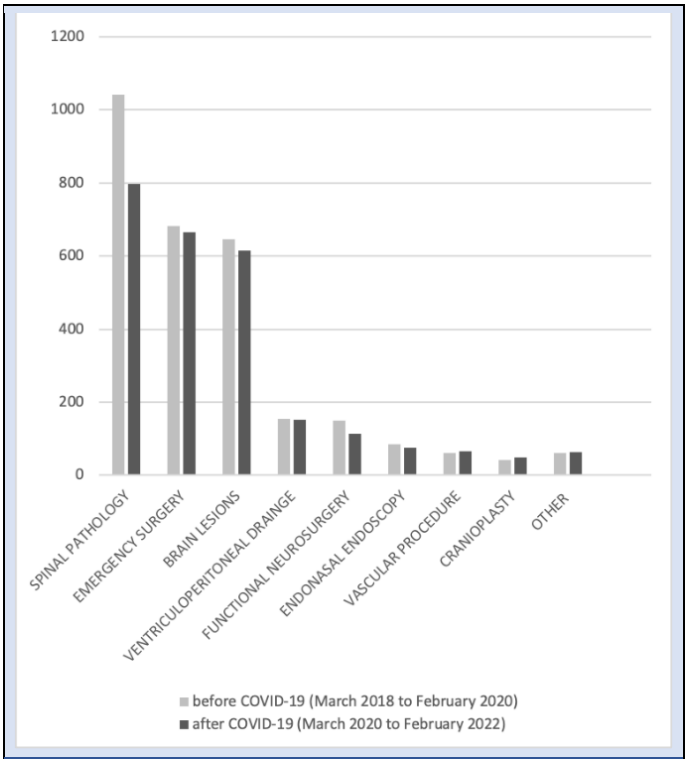

Figure 2. The subgroups of neurosurgical procedures before and after the COVID-19 pandemic.

Overall, we recorded an 11.2% (n=327) decrease in all neurosurgical procedures in two years after COVID-19 was declared compared to two years before. A decrease in elective procedures was 13.9% (n=311), mostly on account of spinal pathology procedures (245, 23.5%), functional neurosurgical procedures (37, 24.7%), endonasal endoscopy procedures (11, 12.8%) and brain lesions (31, 4.8%). The decrease in emergency procedures was insignificant and amounted to a 2.4% drop (n=16) (***Table 1***).

The management protocol in the early phase of the COVID-19 pandemic

The first step included the evaluation and screening of neurosurgical patients who presented for neurosurgical treatment or assessment. In the early phase of the pandemic, the number of positive COVID-19 patients was low. General public measures were set in place: lockdown and limitation of the admission for all urgent patients exclusively through the emergency ward. These patients were managed immediately according to the underlying pathology and simultaneously screened for potential risk of contracting COVID-19 with a nasopharyngeal swab for the rapid antigen test (RAT) initially and then with a nasal swab for the rapid transcriptase-polymerase chain reaction (RT-PCR) test. Imaging, preoperative preparation, and surgical and early postoperative care were performed with all necessary protective measures in positive patients and in those requiring emergent surgical treatment whose COVID-9 status was unknown or unconfirmed at the time of admission. A special isolation area was established where urgent patients were admitted for testing. All emergency, supportive and intensive care hardware was at hand. This area was isolated with no connection to other hospital areas. The initial rapid screening protocol for every patient included body temperature measurement and a detailed COVID-19 screening questionnaire in awake patients. The questionnaire was also administered to patient relatives and attendants, especially in the instances of non-conscious patients, in addition to protective measures such as hand sanitization and face masks.

Elective patients and those requiring non-urgent transfer from other hospitals were screened for potential risk of contracting COVID-19 with the RT- PCR test. They have been provisionally accommodated in so-called transitional zones or specially established holding areas where RT-PCR was performed before surgery. After the test result became available, they were transferred to a regular ward to avoid the potential spread of the virus.

Table 1. Comparison of neurosurgical procedures in a two-year period before and after the COVID-19 pandemic was declared..

Neurosurgical procedure type	Number of cases		Decrease in number of cases
	03.2018 - 02.2020	03.2020 - 02.2022	
Spinal pathology	1042	797	245 (23.5%)
Emergency surgery	681	665	16 (2.4%)
Brain lesions	646	615	31 (4.8%)
Ventriculoperitoneal drainage	154	153	1 (0.7%)
Functional neurosurgery	150	113	37 (24.7%)
Endonasal endoscopy	86	75	11 (12.8%)
Vascular procedure	62	65	/
Cranioplasty	41	50	/
Other	62	64	/
Elective surgery	2243	1932	311 (13,9%)
Emergency surgery	681	665	16 (2.4%)
Total	2924	2597	327 (11.2%)

The management protocol in the late phase of the COVID-19 pandemic

In the later phase of the pandemic, the number the cases rose. The management scheme needed to be adjusted. Therefore, all areas and treated patients were divided into three groups: I) red (danger zone, urgent patients), II) grey (transitional, waiting for the zone, elective patients), and III) green (safe zone, elective patients).The red zone was a high-risk area, comprising the patients with confirmed COVID-19 infection and all vital emergencies transported to the general emergency admission department. These patients required an urgent, lifesaving neurosurgical intervention, regardless of the COVID-19 status, and preventive and protective measures were taken during their treatment. The RAT was used for patients requiring immediate surgery on an emergency basis and the RT-PCR test was performed during the operation to accommodate these patients in suitable postoperative hospital areas. These urgent patients were operated on with full personal protective equipment.

The second, grey, group included all non-urgent and elective patients who were admitted to the neurosurgical department for regular treatment. These patients were either vaccinated or tested in advance (before admission) and were accommodated in grey zones to check their COVID-19 status with RT-PCR. When confirmed virus-negative with RT-PCR, they were transferred to green zones.

The green zone was the safe one, which included COVID-19 negative elective patients. All transfers among these designated hospital and department zones were limited also in terms of personnel, material, and equipment.

All emergency patients underwent this protocol and all admitted patients were categorized based on the degree of emergency for intervention. Those with life-threatening emergencies were operated on immediately, regardless of the COVID-19 status, unless confirmed differently. This was a small group, luckily, and included patients with life-threatening neurosurgical emergencies who were already hospitalised on other hospital wards and were certainly COVID-19 negative when their health condition deteriorated.

The inclusion criteria for emergency surgery encompassed: I) all paediatric and adult patients with features of raised intracranial pressure, like stroke, abscess, tumour, brain oedema, subdural or epidural hematoma, deteriorating hydrocephalus; II) all traumatic cases needing observation or emergency operation; III) all spinal compressive myelopathies (both traumatic and non-traumatic), and IV) vascular emergencies: ruptured intracranial aneurysms, ruptured arteriovenous malformations, intracerebral haematomas.

The urgent cases were triaged according to the complexity of the case, the ability of the anaesthesiology team, the availability of surgical instruments, and the availability of postoperative accommodation (COVID-19/non-COVID-19 emergency rooms).

Urgent patients that could be handled with the existing setup were operated on according to the causal pathology and with all protective measures in the dedicated theatres and postoperatively treated in the designated red zone intensive care units until they were ready for discharge.

Patients with no known exposure to COVID-19 and COVID-19 negative patients, namely the elective patients, semi-urgent patients, and those without acute respiratory signs and symptoms with a normal chest radiograph and negative RT-PCR tests, were considered low-risk. Low-risk patients were operated on in a standard (pre-COVID-19) neurosurgical setting and postoperatively treated in green ICUs or the green ward areas.

Discussion

Despite the difficulties the virus spread has caused to the health system, regular medicine had to work continuously and in parallel with the treatment of COVID-19 patients[13-15]. Therefore, the neurosurgical practice had to be organised according to new rules. The flow of patients to health institutions has increased during the pandemic as the COVID-19 patients have joined patients with everyday health problems. This new reality needed to be addressed adequately and effectively[16,17].

The neurosurgical department in Ljubljana is the largest in Slovenia and addresses all neurosurgical pathology. Due to the constant inflow of patients, it was necessary to limit admissions according to the treatment priority. For this purpose, we have implemented a triage system on the level of the outpatient clinic to minimise patient admission and adapt to the new situation. All emergencies were managed without delay.

When comparing the number of neurosurgical interventions in the two years before and after the COVID-19 pandemic, a relatively small decrease was recorded, amounting to 13.9% elective cases. The drop in cases was most prominent in the fields of spinal pathology and functional neurosurgery since these types of procedures were the least urgent because of the non-malignant nature of pathology in general and the lack of tendency for neurological deterioration. The reduction in surgery for spinal pathology is attributed to fewer surgeries for degenerative spinal disorders with pain syndromes. Cases with accompanying neurological deficits or spinal lesions were operated on as soon as possible when appropriate. The number of procedures in other neurosurgical fields was comparable. There was a small decrease in the number of brain and sellar lesions, which might be coincidental or because patients without acute clinical presentation did not reach adequate health services and did not receive timely appropriate diagnostics for some time during the height of the pandemic.

The decrease in the number of elective surgeries during the pandemic was not as drastic as we predicted it would be. One of the reasons for that was the effective COVID-19 screening protocol. Very few surgeries were postponed on account of a patient being SARS-CoV-2 positive because when a patient presented with a positive test on admission, he or she was rescheduled, returned home, and a substitute patient was called in for the procedure. Another contributing factor might also be that during the quieter periods of the pandemic, when medical staff returned to their original posts, more surgeries were performed to make up for the backlog.

With other SARS-CoV-2 positive patients, if possible, conservative treatment was implied or the patient was rescheduled for the surgery until the contagious period expired.

With these measures, the clinical flow of neurosurgical emergencies was optimised so that the patients were not deprived of immediate neurosurgical intervention. We confirmed this by comparing the number of emergency operations two years before and after the COVID-19 pandemic was declared. In comparison, from March 2018 to February 2020, 681 emergency surgeries were performed and from March 2020 to February 2022, the number of surgeries was 665. Despite the limitations and epidemiological situation, we did not record a significant quantitative decrease in neurosurgical emergency surgeries.

With new triage systems on the level of the outpatient clinic, we optimised patient admission. A nonregulated inflow of patients yields a high risk of the transmission of COVID-19 to patients and hospital staff. In general, SARS-CoV-2 positive patients that undergo surgical intervention have a higher complication and mortality rate than SARS-CoV-2 negative patients[18-21]. COVID-19 is not only associated with a high mortality rate, but also puts an enormous strain on the healthcare system. Severe forms of COVID-19 with respiratory failure led to a high rate of intubation, thus creating a shortage of beds in the intensive care units[14-16]. Therefore, the treatment of other critical diseases became even more difficult and limited. From a neurosurgical perspective, the COVID-19 pandemic forced us to consider that regular neurosurgery needed to continue in parallel with the treatment of COVID-19 and balance these aspects of our everyday work.

As mentioned, special precautions were applied for patients with emergent neurosurgical issues that were COVID-19 positive. These patients were operated on immediately in the COVID-19 operation theatre. The intubation was conducted according to a quick protocol by the anaesthesiologist and the assistant nurse. No other staff were present in the operation theatre at the time of intubation. Then, the surgical staff approached and started the procedure. Personal protective equipment was worn at all times, and institutional protective measures were respected. The equipment and material in the operating theatre were reduced to a minimum. During the surgery, the RT-PCR test was performed to help with the postoperative patient accommodation arrangement. When confirmed positive, patients were further treated in the red zones, which encompassed the COVID-19 intensive care units (ICU) and special areas on the neurosurgical ward. RT-PCR tests were conducted every two days. Sometimes, the patients became COVID positive while hospitalised. In these instances, the patients were transferred to red areas and treated there according to their condition. Extubation was done on the operating table since this minimised the risk for cross-infection and provided ample time for proper operating theatre disinfection. Patients were transferred to the ICU when necessary. Those who were medically stable were treated in the red areas of the neurosurgical ward. They were discharged home when appropriate. In addition to regular precautions, such as minimal drilling and abundant irrigation, we tried to avoid the trans-nasal procedures (for hypophyseal tumours) unless urgent indications (apoplexy, loss of vision) were present. Additionally, clear delineation of roles, a disinfection and aeration plan, cross monitoring of all staff members for potential contamination, and reduction of the number of persons in the theatre at a time is advantageous[20-24].

Conclusions

COVID-19 had a vast impact on the healthcare system in Slovenia, including on neurosurgery. We have attempted to ensure medical services run as normally as possible in our department and others. Management protocols were continuously adapted to the course of the pandemic. New and improved strategies to maintain neurosurgical practice during public health emergencies are necessary for the neurosurgical service and healthcare system to run smoothly in the long term and prevent disruptions during future pandemics.

Disclosures

Conflict of Interest: All authors certify that they have no affiliations with or involvement in any organization or entity with any financial interest (such as honoraria; educational grants; participation in speakers' bureaus; membership, employment, consultancies, stock ownership, or other equity interest; and expert testimony or patent-licensing arrangements), or non-financial interest (such as personal or professional relationships, affiliations, knowledge or beliefs) in the subject matter or materials discussed in this manuscript.

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References

1. Haidar MA, Shakkour Z, Reslan MA, Al-Haj N, Chamoun P, Habashy K, et al. SARS-CoV-2 involvement in central nervous system tissue damage. Neural Regen Res. 2022;17(6):1228-1239. doi: 10.4103/1673-5374.327323.
2. Peacock WF, Soto-Ruiz KM, House SL, Cannon CM, Headden G, Tiffany B, et al. Utility of COVID-19 antigen testing in the emergency department. J Am Coll Emerg Physicians Open. 2022;3(1):e12605. doi: 10.1002/emp2.12605.
3. Chams N, Chams S, Badran R, Shams A, Araji A, Raad M, et al. COVID-19: A Multidisciplinary Review. Front Public Health. 2020;8:383. doi: 10.3389/fpubh.2020.00383.
4. Murta V, Villarreal A, Ramos AJ. Severe Acute Respiratory Syndrome Coronavirus 2 Impact on the Central Nervous System: Are Astrocytes and Microglia Main Players or Merely Bystanders? ASN Neuro. 2020;12:1759091420954960. doi: 10.1177/1759091420954960.
5. World Health Organization. WHO Coronavirus (COVID-19) Dashboard (internet). 2022 August (cited 2022 August 24). Available at: https://covid19.who.int (Accessed 22. 11. 2021)
6. Doglietto F, Vezzoli M, Gheza F, Lussardi GL, Domenicucci M, Vecchiarelli L, et al. Factors Associated With Surgical Mortality and Complications Among Patients With and Without Coronavirus Disease 2019 (COVID-19) in Italy. JAMA Surg. 2020;155(8):691-702. doi: 10.1001/jamasurg.2020.2713.
7. Suleyman G, Fadel RA, Malette KM, Hammond C, Abdulla H, Entz A, et al. Clinical Characteristics and Morbidity Associated With Coronavirus Disease 2019 in a Series of Patients in Metropolitan Detroit. JAMA Netw Open. 2020;3(6):e2012270. doi: 10.1001/jamanetworkopen.2020.12270.
8. Clavien PA, Barkun J, de Oliveira ML, Vauthey JN, Dindo D, Schulick RD, et al. The Clavien-Dindo classification of surgical complications: five-year experience. Ann Surg. 2009;250(2):187-96. doi: 10.1097/SLA.0b013e3181b13ca2.
9. Leira EC, Russman AN, Biller J, Brown DL, Bushnell CD, Caso V, et al. Preserving stroke care during the COVID-19 pandemic: Potential issues and solutions. Neurology. 2020;95(3):124-133. doi: 10.1212/WNL.0000000000009713.
10. Arteaga AS, Aguilar LT, González JT, Boza AS, Muñoz-Cruzado VD, Ciuró FP, et al. Impact of frailty in surgical emergencies. A comparison of four frailty scales. Eur J Trauma Emerg Surg. 2021;47(5):1613-1619. doi: 10.1007/s00068-020-01314-3.
11. Rockwood K, Song X, MacKnight C, Bergman H, Hogan DB, McDowell I, et al. A global clinical measure of fitness and frailty in elderly people. CMAJ. 2005;173(5):489-95. doi: 10.1503/cmaj.050051.
12. NIJZ Koronavirus. Available at: https://www.nijz.si/sl/koronavirus-2019-ncov (Accessed 18. 11. 2021).
13. Olson S, Honeybul S, Rosenfeld JV. Considering Futility of Care Decisions in Neurosurgical Practice. World Neurosurg. 2021;156:120-124. doi: 10.1016/j.wneu.2021.09.078.
14. Jaswaney R, Davis A, Cadigan RJ, Waltz M, Brassfield ER, Forcier B, et al. Hospital Policies During COVID-19: An Analysis of Visitor Restrictions. J Public Health Manag Pract. 2022;28(1):E299-E306. doi: 10.1097/PHH.0000000000001320.
15. Nejadghaderi SA, Saghazadeh A, Rezaei N. Health Care Policies and COVID-19 Prevalence: Is There Any Association? Int J Health Serv. 2022;52(1):9-22. doi: 10.1177/0020731421993940.
16. Agyemang K, Rose A, Baig S, Al Salloum L, Osman AA, Steckler F, et al. Neurosurgery in octogenarians during the COVID-19 pandemic: Results from a tertiary care trauma centre. Interdiscip Neurosurg. 2021;26:101357. doi: 10.1016/j.inat.2021.101357.
17. Sander C, Dercks NV, Fehrenbach MK, Wende T, Stehr S, Winkler D, et al. Neurosurgical Care during the COVID-19 Pandemic in Central Germany: A Retrospective Single Center Study of the Second Wave. Int J Environ Res Public Health. 2021;18(22):12034. doi: 10.3390/ijerph182212034.
18. Ozoner B, Gungor A, Hasanov T, Toktas ZO, Kilic T. Neurosurgical Practice During Coronavirus Disease 2019 (COVID-19) Pandemic. World Neurosurg. 2020;140:198-207. doi: 10.1016/j.wneu.2020.05.195.
19. Panciani PP, Saraceno G, Zanin L, Renisi G, Signorini L, Fontanella MM. Letter: COVID-19 Infection Affects Surgical Outcome of Chronic Subdural Hematoma. Neurosurgery. 2020;87(2):E167-E171. doi: 10.1093/neuros/nyaa140.
20. O'Dwyer MJ, Owen HC, Torrance HD. The perioperative immune response. Curr Opin Crit Care. 2015;21(4):336-42. doi: 10.1097/MCC.0000000000000213.
21. Fontanella MM, De Maria L, Zanin L, Saraceno G, Terzi di Bergamo L, Servadei F, Chaurasia B, Olivi A, Vajkoczy P, Schaller K, Cappabianca P, Doglietto F. Neurosurgical Practice During the Severe Acute Respiratory Syndrome Coronavirus 2 (SARS-CoV-2) Pandemic: A Worldwide Survey. World Neurosurg. 2020;139:e818-e826.
22. Berry G, Parsons A, Morgan M, Rickert J, Cho H. A review of methods to reduce the probability of the airborne spread of COVID-19 in ventilation systems and enclosed spaces. Environ Res. 2022;203:111765. doi: 10.1016/j.envres.2021.111765.
23. Malhotra N., Joshi M., Datta R., Bajwa S.J., Mehdiratta L. Indian Society of Anaesthesiologists Advisory and Position Statement regarding COVID-19. Indian J Anaesth. 2020;64:259-263.
24. Matava C.T., Kovatsis P.G., Summers J.L. Pediatric airway management in COVID-19 patients—consensus guidelines from the Society for Pediatric Anesthesia's pediatric difficult intubation collaborative and the Canadian Pediatric Anesthesia Society. Anesth Analg. 2020;131:61-73.

ORIGINAL RESEARCH

Temporomandibular joint syndrome treatment with peripheral nerve stimulation

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Abstract

Introduction: Temporomandibular joint syndrome is defined by a triad of intense joint pain together with restriction of mouth opening and jaw clicking. The objective of this study is to evaluate the efficacy and safety of peripheral nerve stimulation for the treatment of this pathology.

Material and Methods: A retrospective study was conducted. All patients met selection criteria that include prior resistance to medical or surgical treatment and completion of a series of pre-surgical tests. An octopolar electrode was implanted in the affected preauricular region. The results were measured using the Analog Pain Scale, a short questionnaire on pain, improvement of restriction in mouth opening and reduction of analgesic medication.

Results: A total of 10 patients with 14 performed procedures were included. The mean reduction in pain measured by VAS was 86.2% at one month and 79% at one year after surgery. All patients experienced a drastic improvement in pain and its impact according to the Brief Pain Inventory, the mean improvement being 90% at 4 weeks and 82% at one year. There was an improvement in the mean oral opening of 10.14 mm (minimum of 4 and maximum of 13 mm). One case was excluded due to the complication demanding the system removal.

Conclusions: Patients with temporomandibular joint syndrome who do not respond to conventional treatments are ideal candidates for peripheral nerve stimulation, showing improvement in pain, oral restriction, and quality of life with a low percentage of serious complications.

Keywords: temporo-mandibular joint; peripheral electrical stimulation; auriculo-temporal nerve; orofacial pain syndrome

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Introduction

Temporomandibular joint syndrome (TMJS), or Costen syndrome, is a fairly common functional disorder. The overall prevalence of TMJS was approximately 31% in adults/elderly and 11% in children[1], amounting to the second most common cause of musculoskeletal pain. About 33% of the population has at least one TMJS symptom and 3.6 to 7.0% of the population has TMJS with sufficient severity that they desire treatment[2]. One study found average ratings of pain intensity due to TMJS of 4.3 on a 10-point scale, like the averages reported for chest pain and back pain. Studies consistently find that TMJS has a pronounced impact on quality of life.

TMJS may present with a variety of signs and symptoms and is a diagnosis of exclusion, therefore, all other possible diagnoses must be ruled out[3].

This clinical diagnosis is based mainly on neuropathic pain attributed to the atrio-temporal nerve or mandibular branch of the trigeminal nerve, joint clicking upon mouth opening or chewing, and an anterior meniscus dislocation on magnetic resonance imaging (MRI)[4].

Anamnestic details are of utmost importance, and require a complete historical identification of predisposing, initiating, and perpetuating factors[5]. De Leeuw in 2010, proposed the importance of the physical examination in the diagnosis, which consists of palpation of the temporomandibular joint (TMJ), musculature, active recognition of movement and analysis of joint noise when performed by trained professionals[6]. Psychosomatic, social, and emotional factors may prompt the symptoms of TMJS[7].

Although the optimal treatment remains unclear, determining how to manage TMJS patients would yield significant clinical and economic benefit[8]. Most symptoms improve without treatment, but some patients will require semi-invasive therapies and or potentially invasive therapies such as peripheral nerve stimulation[9]. Candidates for neurostimulation are minimal, though neurostimulation may be underutilized[10].

These patients may fail to attain relief with conventional medical and semi-invasive therapies and require a more invasive treatment. We present a study on peripheral nerve stimulation for the treatment of TMJS.

Material and Methods

Patient Selection

A retrospective analysis of a series of patients with temporomandibular pain treated by the authors between January 2018 and January 2021 was performed.

The inclusion criteria in this series were:

1. Severe pain compatible with temporomandibular joint dysfunction.
2. Neuropathic characteristics and negatively affecting the quality of life of the patients
3. Limitation in mouth opening
4. Resistance to other aggressive treatments performed by the Pain Unit or by Maxillofacial Surgery Unit
5. Positive response to a pre-atrial blockade of the affected joint.
6. Follow-up of at least one year.

The exclusion criteria were:

1. Medical and psychological disorders that prevent adequate intervention or monitoring of patients.

All patients presented with temporomandibular joint pain characterized by intense pain in the masticatory muscles, temporomandibular joint, or both. Pain was present at any time of day, even without jaw activity. Patients also had clicks or cracks when moving the jaw. In more than half of the patients, there was mandibular deviation with opening.

One patient was excluded from the analysis due to unpleasant stimulation of the eye on the stimulated side and allodynia. Her treatment was discontinued, therefore not followed afterwards.

Pre-surgical test

A pre-atrial blockade of the affected joint was performed with 5 ml of 2% lidocaine. This test confirms the existence of a peripheral neuropathy with the possibility of responding to neurostimulation[11,12]. Immediate pain relief and increased mouth opening were considered positive.

Procedure

The electrodes were implanted subcutaneously over the preauricular area of the affected joint. A preauricular incision was made up to the superficial fascia with insertion of the electrode subcutaneously under fluoroscopy control. The final location of the electrode was chosen according to the superposition of the electrical paresthesia on the area of pain. Subsequently, the electrode was tunneled towards the pectoral level, connecting it to a generator in a subfascial pocket. An impedance check was performed, and the battery was left off. The stimulation parameters were established by tailoring conventional procedures of the spinal stimulation systems to the patient (***Figures 1 and 2***).

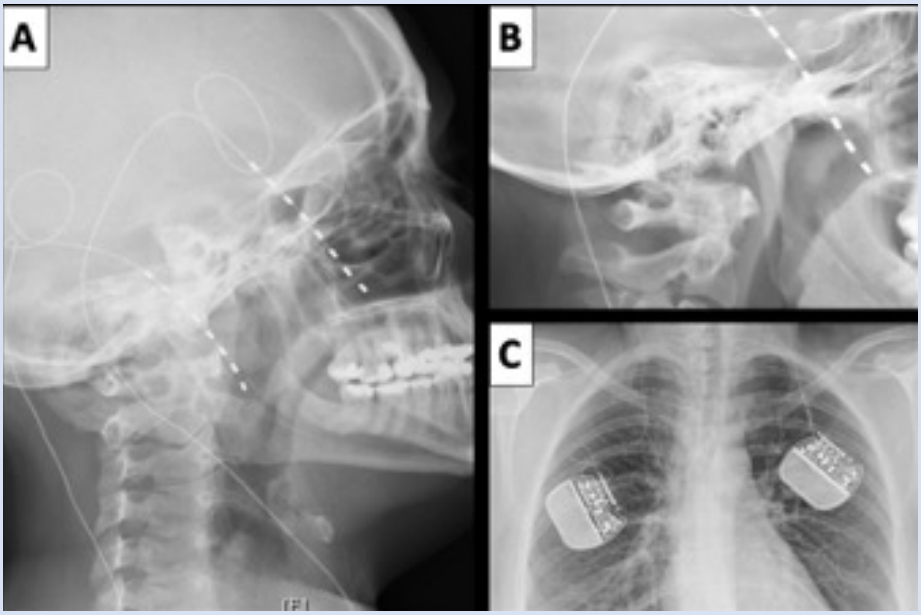

Figure 1. Radiological image showing: **A and B.** the location of the electrodes: at the level of the temporomandibular joint; **C.** the placement of the generators at the subfascial pectoral level

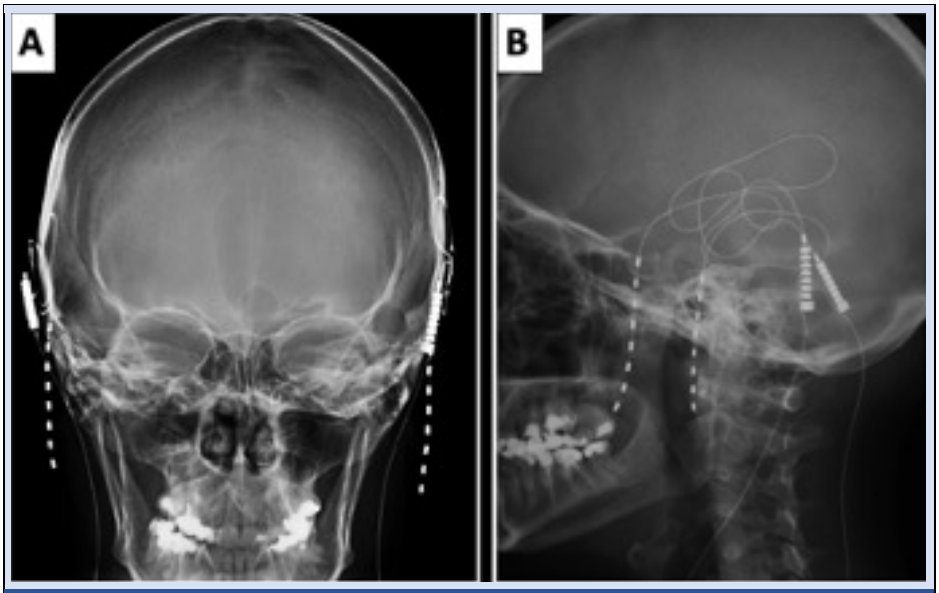

Figure 2. Postoperative radiological image in: **A.** anteroposterior; and **B.** lateral; positions to verify electrode placement.

Outcomes assessment

Outcomes were quantified using the visual analog scale of pain (VAS) and by investigating improvement in mandibular restriction at 1 month, 3 months, 6 months, 9 months, and 1 year following peripheral neurostimulation. Pain intensity, reduction in drugs used for TMJS, the functional status of the patient, and complications were monitored.

The brief questionnaire for the assessment of pain – Brief Pain Inventory (BPI) is a multidimensional pain assessment questionnaire that provides information, not only on the intensity of pain, but also its interference in the daily activities. It was developed by Daut in 1983[13] and was validated in its Spanish version by Badía et al. in 2002[14]. It consists of two dimensions: "pain intensity," with 4 items, and “interference in daily activities," with 7 items. Each item is scored using a numerical scale from 0 (absence of pain / absence of interference in daily life) to 10 (worst pain imaginable / maximum impact on daily life). Based on the results obtained, a summary score is obtained. for each of the two dimensions.

Statistical analysis

Numerical data were presented as median values with ranges (minimum and maximum values), and the absolute values of changes in VAS and BPI values between different assessments were presented as percentage difference.

Results

Demographics

This study involved 10 female patients with a mean age of 41.21 years (range: 31-58 years) who underwent a total of 14 surgeries (8 right, 6 left). One patient was excluded due to the intolerable complication.

They had pain for an average of 7.79 years (range: 2-14 years). TMJS pain was unilateral right in 3 patients (30%) and bilateral in 7 (70%). All had limitation in mouth opening, with an average opening of 19.36 mm (range: 13-25 mm, standard measurement is between 40-60 mm). The number of previous treatments was an average of 6.21 (range: 4-8). These treatments included arthroscopies of the TMJ, injection of botulinum toxin, ozone, and intra-articular injections of lidocaine. No patient presented a significant improvement in pain or mouth opening following the previous treatments. All patients were taking a mean of 4.43 analgesic drugs (range: 3-6 drugs), including at least two opiates.

Treatment Outcomes

All patients attained reduction of pain between 85% and 100% within the first 6 hours of treatment according to the postoperative VAS, performed to adjust postoperative analgesia. The patients had a mean follow-up of 14.36 months (range: 12-28 months).

The initial pre-surgery average VAS was 9.86, including 10 in 12 interventions and 9 in the other 2 surgeries. At the first month after surgery, the mean VAS was 1.36 (range: 0-4). The mean reduction in VAS was 86.2%, and a total of 7 surgeries produced a 100% reduction with a VAS of 0. At 3 months post-surgery, the mean VAS was 2.07 points (0-5 points). The average reduction was 79%, with only in 4 surgeries maintaining a 100% reduction in VAS. At 6 months post-surgery, the mean VAS was 1.79 points (0-4 points). The average reduction was 81.84%. The 100% reduction was maintained in 4 surgeries. At 9 months following surgery, the mean VAS was 2.29 points (0-4 points). The mean reduction was 76.77%. Only 2 surgeries maintained the 100% reduction in VAS. At 1 year post-surgery, the mean VAS was 2.07 points (0-5 points). The average reduction was 79%. A 100% reduction in VAS was maintained in 3 surgeries. ***Figure 3*** demonstrates evolution in VAS over time.

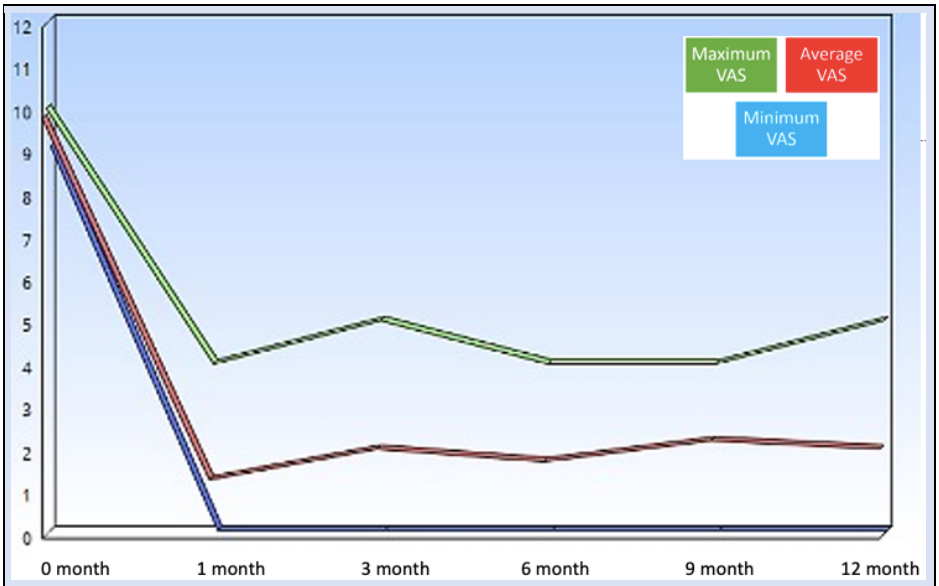

Figure 3. Temporal evolution of the average VAS, maximum VAS, and minimum VAS, demonstrating a clear improvement in pain

All patients experienced a drastic improvement in pain assessment according to the BPI, with a mean improvement of 90% at 1 month, 79.9% at 3 months, 86.6% at 6 months, and 82% at 1 year. Mean mouth opening at 12 months was 29.50 mm (range: 25-38 mm). The mean improvement was 10.14 mm per surgery (range: 4-13 mm). These data are reflected in ***Figure 4***.

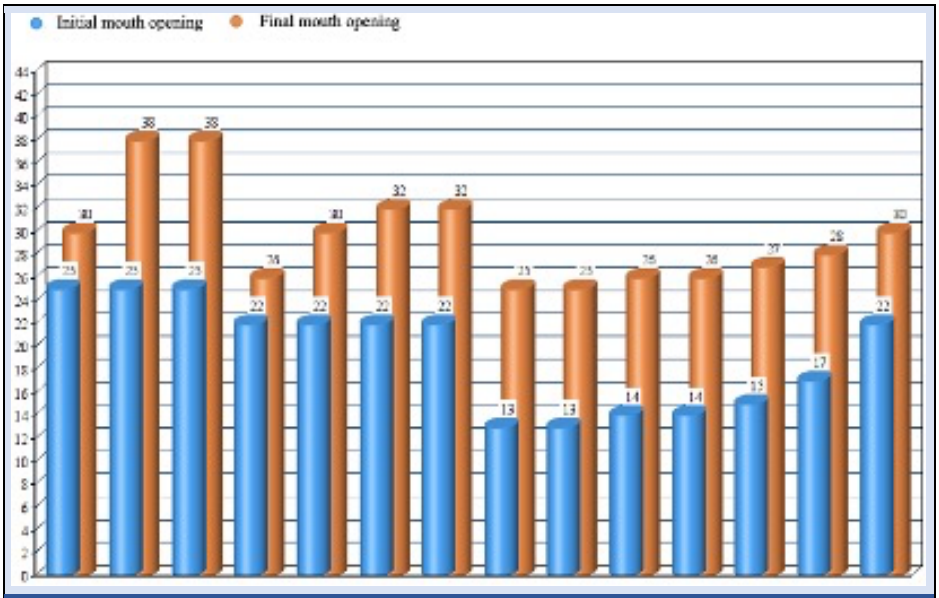

Figure 4. Graph showing the comparison between the initial and final mouth opening for each surgery.

Reduction in the use of analgesic drugs at 12 months was 83.97%% with a mean of 0.72 drugs (range: 0-2). A 100% reduction in analgesic medication was achieved for 7 surgeries. The rest of the surgeries achieved a decrease in analgesic medication that allowed patients to open their mouths, yawn, and chew without pain. All patients normalized their physical activity and sleep.

Complications

There were 2 complications, in one, the patient experienced unpleasant stimulation of the eye and allodynia on the stimulated side, leading to the exclusion from this study analysis, as the stimulation was discontinued within days after implantation. In other patient, breakage of the electrode occurred in at the level of the trajectory 3 months after surgery. It was reimplanted, with no further complications afterwards.

Discussion

Temporomandibular joint dysfunction was first described in 1943 by the Belgian otolaryngologist Dr. James Costen. Drawing on 11 cases, he was the first to suggest that changes in dental conditions were responsible for various otological symptoms. He found cases with symptoms in the region of the TMJ, such as pain of musculoskeletal origin, crackles, otological symptoms such as tinnitus, difficulty opening the mouth, as well as significant unilateral headache[15].

The etiology is multifactorial. All the factors involved can be modulatory or triggers. The most important factors are excessive tension of the jaw muscles causing limited movement of the joint; poor alignment between the upper and lower teeth, resulting in imbalanced movement of the jaw joint; abnormal position or displacement of the jaw joint or cartilage disc within the joint; and jaw pathologies. Possible jaw pathologies include condylar alterations, congenital defects, acromegaly, trauma or dislocation, inflammation or infection of the joint, and bone tumors[16]. A loss of teeth, poorly adapted prostheses, parafunctional habits such as bruxism or nail biting, postural alteration of the jaw and neck, and psychological conditions cause an increase in local muscle activity, leading to spasms and fatigue of the TMJ region[10,16]. Common risk factors are female sex, young adulthood (30-50 years), bruxism, use of very tight dentures, and the presence of other pathologies such as fibromyalgia, stress and arthritis[17].

TMJS presents with very intense pain in TMJ or jaw; extension to one side of the scalp, nape or neck; worsened by chewing, yawning, or talking too much; temporo-mandibular stiffness; difficulty opening the mouth or chewing; popping and cracking joints sensation of closing or brief hooking of the jaw when trying to open or close it; and sensation of muffled hearing, tinnitus, or vertigo. Additionally, diagnosis of TMJS is challenging because its nonspecific and variable symptoms, multidisciplinary workup required, and lack of knowledge among medical professionals.

Conservative medical treatment should always be the first option. This includes administration of a soft diet, avoidance chewing gum, excitants and tobacco, use of dental protector to relax the jaw muscles, and physical therapy. Analgesics, anxiolytics, muscle relaxants or antidepressants are also used[18]. Some patients will be candidates for semi-invasive therapies, such as occlusal adjustment, orthodontics, electrotherapy, botulinum toxin, laser therapy, drug treatment, acupuncture, cryotherapy, and heat therapy[19].

Surgical procedures are considered the last resort. In recent years, the peripheral nerve stimulator has become a very useful surgical resource given its medium and long-term efficacy and low rate of serious complications. Furthermore, unlike other invasive treatments, it is a reversible treatment as it is based simply on stimulation of nerves contributing to paresthesia, rather than destruction of lesions[20]. It was first described in 1967 by Wall and Sweet[21], who found that peripheral neurostimulation produced hypoesthesia and analgesia distal to the stimulated point. They established that the main indication for this procedure is the presence of neuropathic pain[22].

Peripheral nerve stimulation using electrodes is commonly accepted in other pathologies such as headache, facial neuralgia, chronic low back pain, pelvic and perineal pain[23], migraine[24], cluster headache, trigeminal neuralgia[25], postherpetic neuralgia, and post-surgery groin pain[26]. The literature supports its use for neuropathic pain with an efficacy of at least 50% improvement compared to baseline. Few centers perform peripheral nerve stimulation for TMJS. A study of 6 cases found high improvement in pain with few complications limited to rupture of the electrode and unpleasant stimulation of the facial nerve with retraction of the mouth corner[27]. Our study corroborates these findings. However, both series are too small to determine factors associated with efficacy and clarify why some patients, but not others, respond. It is possible that the duration of the disease, which produces nerve involvement in the long term, may modulate the efficacy of peripheral nerve stimulation for TMJS, as with trigeminal neuralgia[28].

Conclusions

Pain secondary to TMJS is a complex and multidisciplinary problem leading to a significant deterioration in the quality of life. Patients who fail to attain relief with conservative and semi-invasive treatments may benefit from peripheral neurostimulation, with a high degree of pain relief and low complication rate. Future studies with larger cohorts will be necessary to validate the findings in this study.

Disclosures

Conflict of Interest: All authors certify that they have no affiliations with or involvement in any organization or entity with any financial interest (such as honoraria; educational grants; participation in speakers' bureaus; membership, employment, consultancies, stock ownership, or other equity interest; and expert testimony or patent-licensing arrangements), or non-financial interest (such as personal or professional relationships, affiliations, knowledge or beliefs) in the subject matter or materials discussed in this manuscript.

Ethical approval: All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee (name of institute/committee) and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed consent: Informed consent was obtained from all individual participants included in the study.

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References

1. Valesan LF, Da-Cas CD, Réus JC, Denardin ACS, Garanhani RR, Bonotto D, Januzzi E, de Souza BDM. Prevalence of temporomandibular joint disorders: a systematic review and meta-analysis. Clin Oral Investig. 2021 Feb;25(2):441-453. doi: 10.1007/s00784-020-03710-w.
2. Velly AM, Anderson GC, Look JO, Riley JL, Rindal DB, Johnson K, Wang Q, Fricton J, Huff K, Ohrbach R, Gilbert GH, Schiffman E; National Dental Practice-Based Research Network Collaborative Group. Management of painful temporomandibular disorders: Methods and overview of The National Dental Practice-Based Research Network prospective cohort study. J Am Dent Assoc 2022;153(2):144-157.
3. Dibbets JM, van der Weele LT. Prevalence of TMJ symptoms and X-ray findings. Eur J Orthod. 1989;11(1):31-36. doi:10.1093/oxfordjournals.ejo.a035961
4. Maglione H, Fernandez Soetbeer G, Allende G. Craniomandibular dysfunction: frequency and forms of presentation in a population of young adults and their demand for care. Research Cabinet. Univ J.F. Kennedy. Rev CAO 2006; 199:32-42
5. Fricton JR, Olsen T. Predictors of outcome for treatment of temporomandibular disorders. J Orofac Pain. 1996;10(1):54-65.
6. De Leeuw R (Ed.) Orofacial Pain: Guideliness for Assessment, Diagnosis, and Management American Academy of Orofacial Pain AAOP . 4th ed. London: Quintessence Publishing Co, Inc., 2008; 8: 167-170. www.aaop.org
7. Becker IM. Occlusion as a causative factor in TMD. Scientific basis to occlusal therapy. N Y State Dent J. 1995;61(9):54-57.
8. Tchivileva IE, Hadgraft H, Lim PF, Di Giosia M, Ribeiro-Dasilva M, Campbell JH, Willis J, James R, Herman-Giddens M, Fillingim RB, Ohrbach R, Arbes SJ Jr, Slade GD. Efficacy and safety of propranolol for treatment of temporomandibular disorder pain: a randomized, placebo-controlled clinical trial. Pain. 2020 Aug;161(8):1755-1767. doi: 10.1097/j.pain.0000000000001882.
9. Buescher JJ. Temporomandibular joint disorders. Am Fam Physician. 2007 Nov 15;76(10):1477-82.
10. Drangsholt M, Le Resche L,. Temporomandibular disorder pain. In:CrombieIK,Croft PR, Linton SJ, LeResche L, VonKorff(eds). Epidemiology of Pain. Seattle: IASP Press, 2004
11. Parate LH, Tejesh CA, Geetha CR, Mohan CV. Peripheral nerve stimulator-guided mandibular nerve block: A report of three cases. Saudi J Anaesth. 2016 Oct-Dec;10(4):491-492. doi: 10.4103/1658-354X.179126.
12. Kalladka M, Shastry S, Thondebhavi M, Quek S, Singh S, Khan J.Nerve blocks in the management of acute temporomandibular disorder emergencies—a narrative review. J Oral Maxillofac Anesth 2022;1. 5 | https://dx.doi.org/10.210
13. Daut RL, Cleeland CS, Flanery RC. Development of the Wisconsin Brief Pain Questionnaire to assess pain in cancer and other diseases. Pain. 1983;17(2):197-210. doi:10.1016/0304-3959(83)90143-4
14. Badia X, Muriel C, Gracia A, et al. Validación española del cuestionario Brief Pain Inventory en pacientes con dolor de causa neoplásica [Validation of the Spanish version of the Brief Pain Inventory in patients with oncological pain]. Med Clin (Barc). 2003;120(2):52-59. doi:10.1016/s0025-7753(03)73601-x
15. Costen JB. A syndrome of ear and sinus symptoms dependent upon disturbed function of the temporomandibular joint. 1934. Ann Otol Rhinol Laryngol. 1997;106(10 Pt 1):805-819. doi:10.1177/000348949710601002
16. Scrivani SJ, Keith DA, Kaban LB. Temporomandibular disorders. N Engl J Med. 2008;359(25):2693-2705. doi:10.1056/NEJMra0802472
17. Phillips JM, Gatchel RJ, Wesley AL, Ellis E 3rd. Clinical implications of sex in acute temporomandibular disorders. J Am Dent Assoc. 2001;132(1):49-57. doi:10.14219/jada.archive.2001.0025
18. Simons DC, Travell JG. Myofascial pain and dysfunction: The trigger point manual. Vol 1. Upper Half of the Body. 2nd Ed. Atlanta, GA: Emory University. 1998.
19. Michelotti A, Iodice G. The role of orthodontics in temporomandibular disorders. J Oral Rehabil. 2010;37(6):411-429. doi:10.1111/j.1365-2842.2010.02087.x
20. Franco Gay ML, López Alarcón MD. Estimulación periférica & subcutánea. Indicaciones. En. Guía Práctica de la neuroestimulación. Grupo español de Neuromodulación. Ed: ESRA-España. Living & Working 2009; Cap.3: p.15-9.
21. Wall PD, Sweet WH. Temporary abolition of pain in man. Science. 1967;155(3758):108-109. doi:10.1126/science.155.3758.108
22. Meglio M, Cioni B, Rossi GF. Spinal cord stimulation in management of chronic pain. A 9-year experience. J Neurosurg. 1989;70(4):519-524. doi:10.3171/jns.1989.70.4.0519
23. Weiner RI. Peripheral nerve stimulation. In: Burchiel KJ (editor). Surgical Management of Pain. New York. Thieme Medical Publishers, Inc 2002. p. 498-504.
24. Brewer AC, Trentman TL, Ivancic MG, et al. Long-term outcome in occipital nerve stimulation patients with medically intractable primary headache disorders. Neuromodulation. 2013;16(6):557-564. doi:10.1111/j.1525-1403.2012.00490.x
25. Lenchig S, Cohen J, Patin D. A minimally invasive surgical technique for the treatment of posttraumatic trigeminal neuropathic pain with peripheral nerve stimulation. Pain Physician. 2012;15(5):E725-E732.
26. Rosendal F, Moir L, de Pennington N, Green AL, Aziz TZ. Successful treatment of testicular pain with peripheral nerve stimulation of the cutaneous branch of the ilioinguinal and genital branch of the genitofemoral nerves. Neuromodulation. 2013;16(2):121-124. doi:10.1111/j.1525-1403.2011.00421.x
27. Rodriguez-Lopez MJ, Fernandez-Baena M, Aldaya-Valverde C. Management of pain secondary to temporomandibular joint syndrome with peripheral nerve stimulation. Pain Physician. 2015;18(2):E229-E236
28. Shi J, Qian Y, Han W, Dong B, Mao Y, Cao J, Guan W, Zhou Q. Risk Factors for Outcomes After Microvascular Decompression for Trigeminal Neuralgia. World Neurosurg. 2020 Apr;136:e559-e566. doi: 10.1016/j.wneu.2020.01.082.

LITERATURE REVIEW AND A SERIES OF CASES

A monocenter retrospective study of the surgical outcomes of adult pilocytic astrocytoma: a small case series and review of the literature

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Abstract

Introduction: The peak age for the development of pilocytic astrocytoma (PA), a type of benign cerebellar tumor, is between 10 and 20 years. Adult PA is extremely rare, and consequently, very little is understood about its characteristics.

Methods: We retrospectively reviewed the records of patients older than 18 years with pathologically proven PA who had surgery to remove the tumor between January 2010 and January 2020 and were followed until January 2022.

Results: Although 32 cases were initially flagged as PA, we included 4 patients (2 male and 2 female) with adult PA. The average age of a male patient at diagnosis was 26.75 years old, and there was no mortality or recurrence. The mean age of female patients at diagnosis was 25 years old. One female was still living after the follow-up period ended. The cause of death in one female patient was unrelated to tumor. Women had a median follow-up of 36 months, and their mean overall survival was 42 months.

Conclusion: PA in adults acts differently than in children. The extent of surgical resection and the location of the tumor influenced the prognosis. When possible, total resection should be the primary treatment, as it promotes good survival rates and low recurrence risk.

Keywords: astrocytoma; pilocytic astrocytoma; glioma; extent of resection

DOI: https://doi.org/10.55005/v3i1.5

Introduction

According to the World Health Organization (WHO), pilocytic astrocytoma (PA) is a grade I tumor with a peak incidence in the first two decades of life and no gender bias. The cerebellum is the typical site of development for PA[1]. Adults PA, however, are extremely elusive[2].

Compact regions of bipolar astrocytes linked with Rosenthal fibers, protoplasmic astrocytes, microcysts, and eosinophilic granular masses are found on histology[1]. The benign and slow-growing nature of PA means that patients have a relatively good prognosis.

Surgery is the treatment of choice since it is curative in cases with complete resection. There is a wide range of reported survival rates, from 86% to 100% five years after surgery, and gross complete resection leads to improved survival. However, poor outcomes have been linked to incomplete resection and tumors in surgically inaccessible areas[3–5].

Even worse outcomes were seen in a recent study of PA patients in adulthood[2]. However, few studies have focused on adult PA patients, rendering the effect of age on prognosis unclear[2,6,7]. During the last decade, we have recorded 4 cases of PA in adults. The goal of this study was to examine the surgical outcomes of PA in adults and to identify potential influencing factors.

Materials and Methods

The Center of Neurosurgery and Neurology cared for 32 patients with PA between 2010 and 2020. We did not include 28 patients in our study because they were either under 18 or had missing records. Four patients met our inclusion criteria and were examined retrospectively. We performed surgery on adults diagnosed with PA (astrocytoma grade I as determined via histopathology). Up until January 2022, their clinical data were reviewed. Patient demographics, clinical history, radiographic findings, operational particulars, tumor features, and pathology information were extracted. There was a general consensus that all cases presented with neurological complaints.

Preoperative magnetic resonance imaging (MRI) with and without gadolinium contrast enhancement was performed on all patients meeting these criteria. An MRI scan was performed within 72 hours after surgery in all operated cases. Based on pre-op volume (X) and post-op volume (Y), Extent of Resection (EOR) was calculated using the formula X-Y/X*100%. After obtaining EOR%, we classified, more than 99% as gross total resection (GTR), 91-99% as near-total resection (NTR), 70-90% as sub-total resection (STR), less than 70% as partial resection and as biopsy. As part of the follow-up for each patient, a series of MRI scans were acquired.

Results

A total of 4 patients, including 2 males and 2 females, were included. Mean age of male at diagnosis was 26.75 years. At the end of follow-up, there was no male mortality and there was no recurrence. Mean age of females at diagnosis was 25.5 years. At the end of follow up period, 1 female was alive. Mean OS in females was 42 months, and there was recurrence in 1 female patient. According to Burkhard et al., while there is a minor preference for the supratentorial space in adults (55%), the cerebellum is the most common site for tumors in children (67%)[3]. PA occurred in cerebellum in 1 case and in the brainstem in 3 cases. Only data from resectable adult patients were analyzed based on preoperative imaging. Due to tumor location, a gross total resection (GTR) was attained in 1/4 of patients, and an NTR was achieved in 1/4 of patients. However, 2/4 patients attained partial resection.

Patients were followed for a median of 36 months after hospital release, with a range of 2 months to 144 months. From a total of 4 adults diagnosed with PA, 3 were clinically stable or improved after 42 months on average and 1 had passed away in that time. One of four patients experienced a recurrence. We anticipate that the survival rate reflects the overall positive prognosis of this tumor type, as demonstrated by previous studies.

Discussion

Most commonly found in children and young adults, PA are slow-growing, benign, low-grade glioma tumors with a clear boundary[1,8]. PA is the most frequent glioma in children, comprising as many as 25% of all brain tumors removed through neurosurgical practice. However, PA is uncommon in adults, accounting for only 2.3% of all brain tumors[2], leading to a lack of knowledge regarding their features. This ten-year retrospective study discusses the demographics and treatment outcomes of a cohort of adult patients with PA treated at the Clinical Center of Neurosurgery and Neurology.

Recently, cases of PA in patients older than 30 years have been noted. A case was detected by Rossi et al. in the hypophysis of a man aged 40, and Kehler reported four cases in people aged 40 and up, albeit the specific details of these cases were not specified[9,10]. Of the 37 cases discussed by Lee et al., only one was older than 20 years[11]. Burkhard et al. reported a recent series of 55 participants, of whom 11 were 30 or older[3]. Only 4 of our 32 patients with PA were 18 or older.

While PA can appear anywhere along the neuraxis, it is most often found in midline structures such the cerebellum, optic chiasm, and brainstem. The cerebellum is the most common origin of PA, though in our series 1 case was located in the cerebellum and 3 were located in the brainstem. Patients presenting with PA have varying clinical manifestations depending on the size and site of the lesion. Our study found that the majority of people with posterior fossa PA also had difficulties walking due to symptoms of increased intracranial pressure, such as headaches, vomiting, and impaired vision.

Multiple studies have demonstrated that the location and extent to which a tumor was excised after surgery significantly impact prognosis. A univariable Cox proportional hazards regression analysis of 865 adult patients with PA from the USA National Cancer Institute Surveillance, Epidemiology, and End Results Program database indicated that gross total resection reduced the risk of death by a factor of 0.2 compared to subtotal resection or biopsy[12]. Similarly, Stüer et al. found that individuals whose tumors were only partially removed had a fourfold higher recurrence rate than those whose tumors were removed completely[2]. Total tumor removal is not only related to improved outcomes, but also curative. The patient in our study who underwent a total tumor removal had no tumor recurrence or death. Optic chiasm and brainstem PAs are associated with poorer outcomes due to the increased difficulty of access and complete excision. In our series, one patient with brainstem PA had a recurrence of their tumor, while another patient with brainstem PA passed away during follow-up.

Depending on the study, the prognosis for PA in adults can range from excellent to poor[2,3,8,12]. This is due to conflicting data showing either greater survival rates or worse prognoses and a higher likelihood of tumor recurrence following surgical excision. In the National Cancer Institute database study, survival rates decreased dramatically with age, from 96.5% at 5–19 to 52.9% at over 60 years of age[12].

Among 44 adult patients in a retrospective study, 5-year survival was 87% and progression-free survival (PFS) was 72%[2]. Brown et al. found PFS and OS rates of 95% after 5 years in a prospective study comprising 20 patients[12].

Based on the data we gathered, PA in adults may not be completely benign. One of four patients in our study experienced tumor recurrence and one died during the study period, with an overall survival rate of 42 months. The discrepancy between this study's findings and those of other investigations may be attributable, in part, to the small sample size of adult patients studied in this study.

Conclusions

Adult PA is an uncommon tumor with a similar clinical presentation to that of childhood/adolescence. The standard of care appears to be surgical resection. The prognosis is favorable if entire resection can be performed. To confirm these preliminary findings, further studies are needed. Extent of resection and tumor site were shown to influence prognosis. We recommend careful monitoring and follow-up for adult PA patients.

Disclosures

Conflict of Interest: All authors certify that they have no affiliations with or involvement in any organization or entity with any financial interest (such as honoraria; educational grants; participation in speakers' bureaus; membership, employment, consultancies, stock ownership, or other equity interest; and expert testimony or patent-licensing arrangements), or non-financial interest (such as personal or professional relationships, affiliations, knowledge or beliefs) in the subject matter or materials discussed in this manuscript.

Ethical approval: All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. This study was approved by the Research Ethics Committee, Faculty of Medicine, Neurosurgery Department, Uzhhorod National University.

Informed consent: Informed consent was obtained from all individual participants included in the study.

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References

1. Burger PC, Scheithauer BW, Paulus W et al. Pilocytic Astrocytoma. In Kleihuesb P, Cavanee WK, editors. Pathology and genetics of tumours of the central nervous system. Lyon: IARC Press, 2000: 45-51
2. Stüer C, Vilz B, Majores M, et al. Frequent recurrence and progression in pilocytic astrocytoma in adults. Cancer 2007;110:2799–808 doi: 10.1002/cncr.23148
3. Burkhard C, Di Patre PL, Schüler D, et al. A population-based study of the incidence and survival rates in patients with pilocytic astrocytoma. J Neurosurg 2003; 98:1170–4 doi: 10.3171/jns.2003.98.6.1170
4. Forsyth PA, Shaw EG, Scheithauer BW, et al. Supratentorial pilocytic astrocytomas: a clinicopathologic, prognostic, and flow cytometric study of 51 patients. Cancer 1993; 72:1335–42 doi: 10.1002/1097-0142(19930815)72:4<1335::aid-cncr2820720431>3.0.co;2-e
5. Kayama T, Tominaga T, Yoshimoto T. Management of pilocytic astrocytoma. Neurosurg Rev 1996; 19:217–20 doi: 10.1007/BF00314833
6. Bell D, Chitnavis BP, Al-Sarraj S, et al. Pilocytic astrocytoma of the adult-clinical features, radiological features and management. Br J Neurosurg 2004;18:613–6 doi: 10.1080/02688690400022896
7. Brown PD, Buckner JC, O'Fallon JR, et al. Adult patients with supratentorial pilocytic astrocytomas: a prospective multicenter clinical trial. Int J Radiat Oncol Biol Phys 2004; 58:1153–60 doi: 10.1016/j.ijrobp.2003.09.020
8. Pagni CA, Giordana MT, Cavanero S. Benign recurrence of a pilocytic cerebellar astrocytoma 36 years after radical removal: Case Report. Neurosurgery 1991; 28:606-9 doi: 10.1097/00006123-199104000-00023
9. Rossie MI, Bevan JS, Esiri MM et al. Pituicytoma (pilocytic astrocytoma). Case Report. J Neurosurg 1987; 67:768–72 doi: 10.3171/jns.1987.67.5.0768
10. Kehler U, Arnold H, Muller H. Long term follow up of infratentorial pilocytic astrocytomas. Neurosurg Rev 1990; 13: 315-20 doi: 10.1007/BF00346373
11. Lee YY, Van Tassel P, Bruner JM, Moser RP, Share JC. Juvenile pilocytic astrocytomas: CT and MR characteristics. AJR 1989; 152:1263-70 doi: 10.2214/ajr.152.6.1263
12. Johnson DR, Brown PD, Galanis E, et al. Pilocytic astrocytoma survival in adults: analysis of the surveillance, epidemiology, and end results program of the National Cancer Institute. J Neurooncol 2012; 108:187-93 doi: 10.1007/s11060-012-0829-0

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CASE REPORT

Head slippage following displacement of a Mayfield head clamp leading to a unique complication of laryngeal edema in an intubated patient: case report

Tahreem Fatima, MBBS,MRCSEd[1]

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Abstract

Introduction: The Mayfield head clamp (MHC) is a three-pin skull clamp that allows excellent cranial stabilization during head and neck surgery and is the most frequently used head clamp in neurosurgery. In many cases, surgery is performed with complete reliance on the safety of the MHC. However, potentially serious or life-threatening complications, such as scalp lacerations, depressed skull fractures, venous emboli, or extradural hematoma can rarely occur.

We describe a case in which the MHC displaced downwards due to sudden loss of grip during postoperative removal with a brief review of the literature.

Case report: The patient was a 60-year-old male who was scheduled to undergo tumor resection of a recurrent bilateral parasagittal parafalcine meningioma via a bicoronal approach. While removing the MHC, the lower screw stabilizing the arm of the MHC system displaced downwards (when force was applied from both directions) leading to an extension of flexed neck along with jerking of the cervical spine despite hand stabilization of the head in an intubated patient with slight upward displacement of a tightly fixed endotracheal tube.

Conclusion: We emphasize the importance of properly managing and maintaining instruments to prevent fatal injury.

Keywords: Mayfield head clamp; neurosurgery; complication; laryngeal injury

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Introduction

The Mayfield head clamp (MHC) (MAYFIELD™, Ohwa Tsusho Co., Ltd., Tokyo, Japan) and (Ohio Medical Instrument Co., Cincinnati, Ohio) (***Figure 1***) is the most frequently used head clamp system in the field of neurosurgery. In many cases, surgery is performed with complete reliance on the safety of the MHC[1].

However, we experienced an extremely rare case in which the MHC accidentally displaced downwards due to loss of screw grip of this MHC, leading to displacement of the endotracheal tube (ETT) during postoperative removal.

Upward pulling of an externally fixed cuffed ETT secondary to uncontrolled hyperextension of the head might have resulted in larynx mucosa injury.

Figure 1. Mayfield head clamp apparatus.

Case report

The patient was a 60-year-old male who underwent a scheduled resection of a recurrent bilateral parafalcine meningioma via a bicoronal approach.

While removing the three fixed pins of the MHC, the lower screw stabilizing the arm of the MHC system displaced downwards when force was applied from both directions. Fortunately, the patient remained in a stable position and did not sustain an injury to the head or neck. The patient was extubated following removal of the MHC at the conclusion of surgery.

The patient was admitted to the intensive care unit (ICU) shortly after extubation and weaning of general anesthesia. However, the patient developed hoarseness and stridor after arrival in the ICU. After a detailed assessment by the anesthesia team, a plan for reintubation via endotracheal tube with subsequent flexible bronchoscopy was pursued.

During reintubation, marked laryngeal edema was noted with a slight contusion over vocal cords (***Figure 2***).

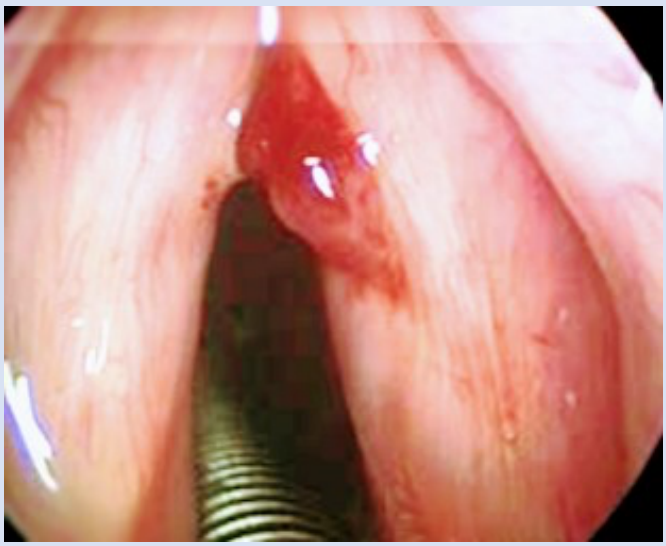

Figure 2. Laryngeal edema with vocal cord injury..

The patient was sedated, and 12 hours following reintubation, developed marked submandibular and supraclavicular emphysema more prominent on the right (***Figure 3***).

Figure 3. Submandibular and supraclavicular emphysema

To decrease swelling, systemic corticosteroid therapy was started along with nebulization. The chest surgery team was consulted. Chest and lung imaging was performed on an emergency basis. No tracheobronchial injury was found. The patient was weaned off the ETT over the course of the next 3 days with subsequent improvement in subcutaneous emphysema. The author experienced an extremely rare case of MHC slippage during removal and emphasizes the importance of properly managing and maintaining the instruments in order to prevent fatal injury.

Discussion

According to the Ohio Medical Instrument Co., the MHC base unit is of a marine grade and highly suited for use in a saline environment[2,3]. The material specifications call for a shear strength of 27,000 pounds per square inch (psi) and a modulus of elasticity of 10.3 million psi. This material is used for when products are placed under sustained loads without the use of torque wrenches to control load forces. However, as described above, although this material is designed for excellent weight bearing and durability, screw locking and unlocking may be unpredictable due factors such surgical environment, material changes over prolonged usage, and length of surgical procedures. Pressure bearings and slight increase of force may result in exponential response of system, as in our case. MHC may be subject to damaging forces due to overuse, particularly in low- and middle-income countries. Screw fatigue exhaustion often results from overuse coupled with a lack of maintenance services. ***Figure 1*** presents arrows indicated where the sudden loss of screw grip of the instrument occurred during the attempt to remove the MHC at the conclusion of surgery. ***Figure 4*** demonstrates a speculated line along which downward displacement of the patient's head occurred due to the weight of the patient's head and the force of gravity. Additionally, the externally fixed ETT was in a relatively insecure position while removing the MHC apparatus and suddenly gave way as the patient's neck pulled on it[4,5].

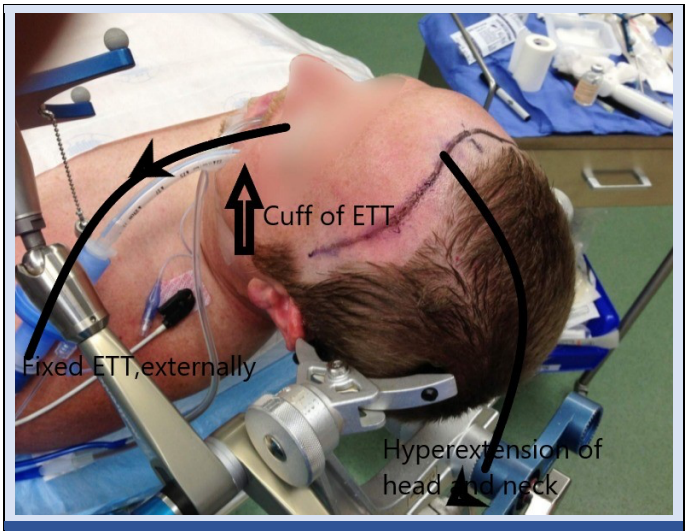

Figure 4. Possible mechanism of injury. ETT - endotracheal tube.

Laryngeal injury was likely due to the normal larynx on preoperative assessment with no documented complications reported by the anesthesia team due to cuff inflation and the lack of subcutaneous emphysema preoperatively. Upon extubation, the patient the patient developed immediate respiratory distress, which ultimately required intubation. Laryngeal edema and a small hematoma were visualized. A total of 12 hours after reintubation, the patient developed subcutaneous emphysema. In the light of witnessing screw grip of the MHC, we searched for and found additional articles reporting ETT displacement related larynx mucosal injuries[6].

During surgery, neurosurgeons rely completely on the safety of the MHC. Without it, the patient's head would not be stabilized. Since this apparatus can cause fatal injury if it breaks during neck or brain surgery, surgeons must fix three MHC pins accurately and with proper pressure[7]. Moreover, surgeons repeatedly confirm immobilization of the connection parts prior to surgery and intraoperatively. It is very important to examine the apparatus thoroughly before each use to prevent unnecessary complications.

The MHC that slipped and displaced was the more commonly used of the two MHCs owned by the hospital. The author had been assisting in surgeries using this MHC for the past 3 years in approximately 500 surgeries.

Only the pins are subjected to sterilization, and the remainder of the device is cleaned but not sterilized. The MHC was always stored in a designated place determined by the operating theater staff.

When the clamp was removed, a downward force was applied, leading to a loss of neck flexion with sudden loss of control due to loosening of lower arm screw. The cervical spine jerked despite head stabilization, leading to an upward displacement of the ETT. The underlying mechanism was probably fatigue related displacement of the screw after slippage.

In a previous report, Taira et al. reported a case in which the MHC broke[3]. The patient's head fell suddenly to the level of the surgeon's knee when operating on a cerebral aneurysm under a microscope, and with povidone-iodine solution was found inside. Chovanes sent a letter to Taira et al. highlighting the importance of carefully inspecting the MHC before every operation as he found signs of imminent break upon reading their paper and inspecting the equipment, he used[1].

Another case reported by Lee et al. described an accidental brakeage of the MHC acute angle arm while installing this system for immobilization of the head and neck[7].

The three clinical cases of MHC breakage, including our case, are summarized in ***Table 1***.

Table 1. Summary of reported Mayfield head clamp malfunctions.

Author, Year	Age/Sex	Procedure	Event time	Problem site	Use period	Broken part replacement	Result
Taira & Tanikawa, 1992	68/F	Aneurysm clipping	During operation	Arm joint broken	8 years (>1200 uses)	Unknown	Uneventful
Lee et al., 2009	58/F	Cervical laminoplasty	During preoperative preparation	Acute angle angle arm broken	10 years (>1000 times)	Other head clamp replacement	Uneventful
Fatima, 2023	60/M	Meningioma resection	During removal	Lower screw displaced	3 years (>500 uses)	N/A	Laryngeal injury

The cases in question occurred most likely due to the fatigue of the equipment. However, the majority of complications occurring in relation to the use of MHC are due skull fractures with or without epidural hematoma, occurring due to the limitless force applied when applying the MHC[8].

Thiys and Menovsky suggested that the skull clamp application requires proper attention to several details and provided a checklist to be used. They also emphasized the role of supervision of an experienced neurosurgeon and the application of the MHC on synthetic or cadaver skulls as an easy and efficient way to practice and understand proper pinning for different cranial approaches[8].

Surgeons and nurses must carefully handle all tools used during surgery to prevent tool-related problems. Damaged and malfunctioning tools should not be used and can lead to surgical failure and intra- and postoperative complications. Safety is of utmost importance when using position fixation instruments[2]. Thus, following operating instructions provided by the manufacturer must be followed during surgery.

The first step in preventing fatal injury is to apply and remove the instrument in the correct position with protection from impact and physical damage. Screws should be regularly checked to detect fatigue failure of the MHC. Also, all cleaning and sterilization procedures should be performed in accordance with the operating instructions provided by the manufacturer.

The surgeon should examine the apparatus thoroughly before each use. Finally, instruments should be subjected to regularly scheduled maintenance by the manufacturer through a separate contract when purchasing the MHC. However, most buyers ignore this aspect in an effort to reduce costs.

Conclusions

Proper management and maintenance of all instruments, and especially those severely relied upon is of utmost importance to prevent potential injury.

Disclosures

Conflict of Interest: All authors certify that they have no affiliations with or involvement in any organization or entity with any financial interest (such as honoraria; educational grants; participation in speakers' bureaus; membership, employment, consultancies, stock ownership, or other equity interest; and expert testimony or patent-licensing arrangements), or non-financial interest (such as personal or professional relationships, affiliations, knowledge or beliefs) in the subject matter or materials discussed in this manuscript.

Ethical approval: All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

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References

1. Chovanes GI. Breaks in Mayfield head holder. J Neurosurg. 1992 Dec;77(6):977. doi: 10.3171/jns.1992.77.6.0977.
2. Day JL. Maintenance of Mayfield head rest. J Neurosurg. 1992 Dec;77(6):977. doi: 10.3171/jns.1992.77.6.0977a.
3. Taira T, Tanikawa T. Breakage of Mayfield head rest. J Neurosurg. 1992 Jul;77(1):160-1. doi: 10.3171/jns.1992.77.1.0160.
4. Atteya MME, Raslan S, Elkallaf M, Soliman MS, AlQalla MA. A novel method for safe and accurate placement of the rocker pins of head immobilization devices utilizing a digital caliber phantom: technical note. *Childs Nerv Syst*. 2019;**35**(9):1599–1602. doi: 10.1007/s00381-019-04216-x
5. Baerts WD, de Lange JJ, Booij LH, Broere G. Complications of the mayfield skull clamp. *Anesthesiology*. 1984;**61**(4):460–461. doi: 10.1097/00000542-198410000-00017
6. Marty-Ané CH, Picard E, Jonquet O, Mary H. Membranous tracheal rupture after endotracheal intubation. Ann Thorac Surg. 1995 Nov;60(5):1367-71. doi: 10.1016/0003-4975(95)00643-Y.
7. Lee TH, Kim SJ, Cho DS. Broken mayfield head clamp. J Korean Neurosurg Soc. 2009 May;45(5):306-8. doi: 10.3340/jkns.2009.45.5.306.
8. Thijs D, Menovsky T. The Mayfield Skull Clamp: A Literature Review of Its Complications and Technical Nuances for Application. World Neurosurg. 2021 Jul;151:102-109. doi: 10.1016/j.wneu.2021.04.081.

CASE REPORT

Appearance and rupture of a dissection aneurysm of the basilar trunk in a SARS-CoV-2 patient

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Abstract

The article describes a clinical case of a dissection aneurysm of the basilar artery in a SARS-COV-2 patient. The patient was treated in the hospital for infectious diseases due to bilateral poly-segmental pneumonia complicating a COVID-19 infection. A focal neurological deficit suddenly appeared and rapidly progressed, including paresis of the abducens nerve, bulbar disorders, and a right-sided hemiparesis. During the course of treatment, the patient underwent CT and CT angiography of the cerebral arteries several times. During the first examination, no pathology of the cerebral vessels was discovered. During the examination 10 days later, a fusiform aneurysm of the basilar artery trunk with a daughter sac was detected. Its rupture led to massive subarachnoid hemorrhage and an unfavorable outcome. The occurrence of primary symptoms in this patient could be due to a violation of the blood flow in the short branches of the basilar trunk in the area of the dissection.

Keywords: SARS-COV-2; COVID-19; stroke; subarachnoid hemorrhage; dissection aneurysm

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Introduction

The effects of the novel coronavirus 2019 (SARS-CoV-2) have not been fully elucidated. It is difficult and not always possible to reliably prove the relationship between an atypical course or outcome of a particular disease with SARS-CoV-2, especially in individual clinical cases or small series of observations. Nevertheless, it is very important to record such clinical cases, accumulate new data, and increase the awareness of colleagues about possible rare complications of SARS-CoV-2.

Clinical cases, series of observations, literature reviews, and several meta-analyses on cerebrovascular complications of SARS-CoV-2 were published in 2020-2021[1-6]. There was an increase in the frequency of stroke, decompensation of encephalopathy, and development of demyelinating diseases attributed to SARS-CoV-2 [2-4,7-9]. According to some reports, this is due to direct damage of the endothelium of cerebral vessels, glial cells and neurons, as well as the development of hypercoagulation and thrombocytopathy caused by the SARS-CoV-2 virus[3,4,9].

In SARS-COV-2 patients hospitalized in infectious diseases hospitals, hemorrhagic stroke develops in 0.2-1.5% of cases[2,3,7,10]. The frequency of subarachnoid hemorrhages (SAH) in patients with SARS-CoV-2 is not reliably known, and in the literature of the last two years, there are only isolated clinical cases and small series of observations[6,7,11,12]. Nawabi et al. (2020) analyzed 18 cases of non-traumatic intracranial hemorrhages (ICH) in SARS-CoV-2 patients, among whom six patients had ICH (33.3%), 11 had SAH (61.1%), and one patient had non–traumatic subdural hematoma (5.6%)[8]. This ratio of different locations of hemorrhages in patients with a new coronavirus infection is unusual, given that in non- SARS-CoV-2 patients, SAH is only seen in 10-15% of cases. Some researchers suggest that SARS-CoV-2 can cause the formation of dissection aneurysms, as well as lead to rupture of existing saccular intracerebral aneurysms[7,13].

This article presents a rare case of a SARS-CoV-2 patient with formation and rupture of a basilar artery dissection aneurysm within a few days of infection.

Case report

A 45-year-old male patient was admitted to the infection diseases hospital on December 18, 2021. He was treated for a SARS-CoV-2 infection complicated by bilateral poly-segmental pneumonia. Since admission, the patient had a severe headache (up to 8 points on a visual analog scale).

To exclude acute cerebral pathology, the patient was examined by a neurologist, who revealed convergent strabismus due to paresis of the left abducens nerve. Multi-slice computerized tomography (MSCT) and MSCT angiography of the brain were performed but it remained unremarkable (***Figure 1***).

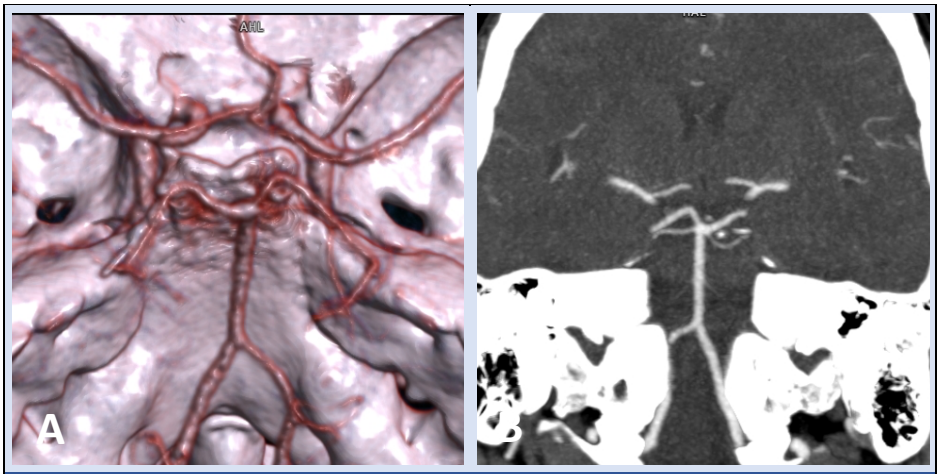

Figure 1. MSCT angiography: A. 3D reconstruction; B. CT angiography of the vertebrobasilar trunk. The basilar artery was unchanged.

January 2, 2022: The patient's headache worsened. There were no finding supporting a diagnosis of SAH. To exclude CNS infection, a lumbar puncture was performed. Cerebrospinal fluid (CSF) pressure was not increased, the color of CSF was transparent, with 4 cells in 1 μl and a protein level of 0.43 g/l.

On January 6th the patient had fever up to 30-38.5°C, leukocytosis up to 15*10^{9}, and C-reactive protein was 145. There was no consciousness disturbance (Glasgow Coma Scale (GCS) 15), he was emotionally labile, with a paresis of the left abducens nerve, and no meningeal signs. CSF from lumbar puncture contained 680 cells in 1 μl and 0.56 g/l proteins. Meningoencephalitis was diagnosed and antibiotics were prescribed.

Next morning, the patient complained of slurred speech and weakness in his right arm and leg. Neurological examination noted GCS 12, palpebral fissures asymmetry D<S, dysarthria, paresis of the left abducens nerve, and a moderate right-sided hemiparesis. Since a stroke was suspected, MSCT was repeated but detected no structural pathology of the brain, to diagnose an acute meningoencephalitis with oculomotor disorders (paresis of the left VI CN). A differential diagnosis of vertebrobasilar stroke was considered.

In the evening, worsening of hemiparesis to hemiplegia, reduced pharyngeal reflex, and dysphagia were noted. Repeated MSCT and MSCT-angiography revealed a fusiform aneurysm of the basilar artery with a daughter sac directed to the left, without structural brain pathology (***Figure 2***).

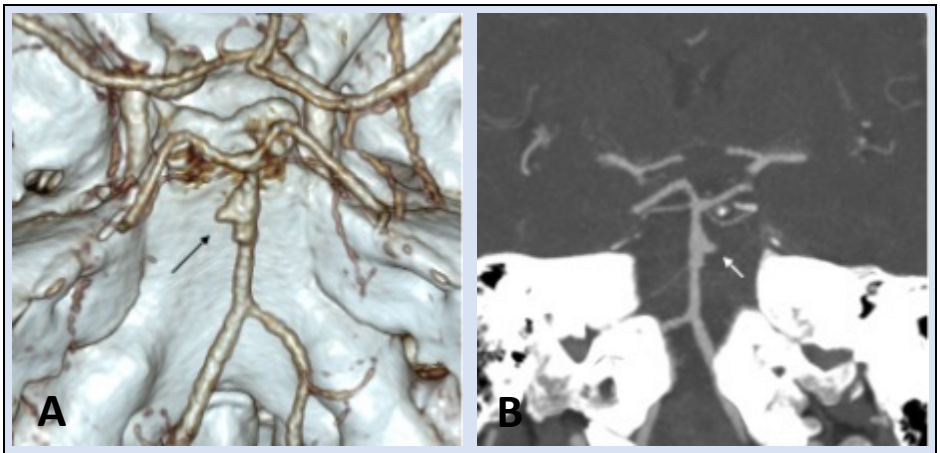

Figure 1. MSCT angiography, 10 days after the initial. A. 3D reconstruction; B. fusiform aneurysm of the basilar artery with a daughter sac directed to the left.

The patient was seen by a neurosurgeon. Cerebral digital subtraction angiography (DSA) was planned to clarify the possibility of endovascular treatment of the aneurysm.

On the morning of January 8, 2022, the patient's condition worsened, with deterioration of consciousness to coma (GCS 6). The patient was moved to the intensive care unit, intubated, and artificial ventilation of the lungs was started. A CT scan of the brain was performed immediately. A massive SAH (Fisher III) was detected (***Figures 3A and 3B***) and MSCT angiography showed an increase in size of the fusiform aneurysm of the basilar artery (***Figures 3C and 3D***).

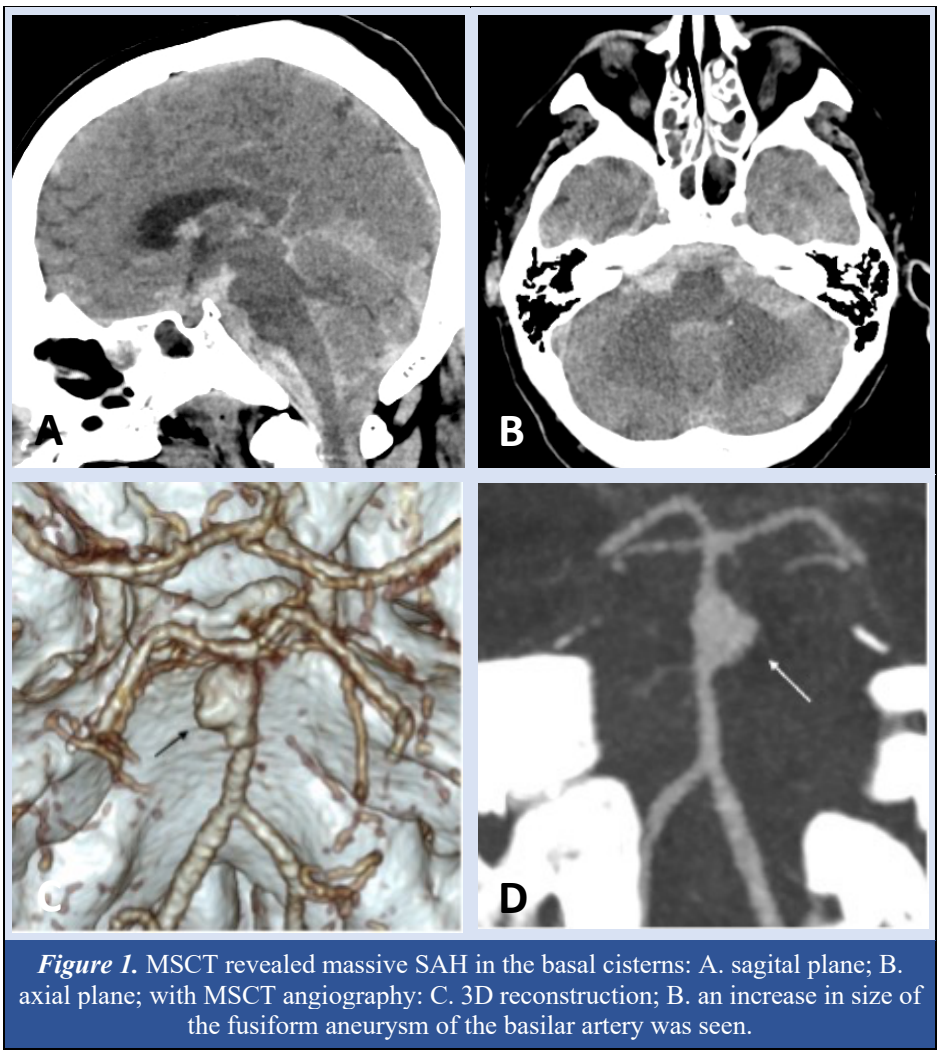

Figure 1. MSCT revealed massive SAH in the basal cisterns: A. sagital plane; B. axial plane; with MSCT angiography: C. 3D reconstruction; B. an increase in size of the fusiform aneurysm of the basilar artery was seen.

Repeated examination by a neurosurgeon, taking into account the severity of the condition (Hunt-Hess V), found no indication for surgical treatment.

January 9, 2022: The patient's condition was terminal, demonstrating coma III, muscular atony, and areflexia. Cardiac arrest was recorded despite increasing doses of cardiotonic drugs. Resuscitation was conducted for 30 minutes without effect followed by a lethal outcome.

Discussion

Analyzing this clinical case, it is possible to suggest the following sequence of pathogenetic events: the COVID-19 virus, tropic to the endothelium, caused damage to the cerebral vessels, with greater involvement of the basilar artery and inflammation of its wall, which led to the destruction of the endothelium and the internal elastic membrane, triggering dissection. Against the background of progressive dissection of the wall of basilar artery, there was a consistent violation of blood supply in the perforating arteries that supply the brainstem, accompanied by the appearance and increase of focal neurological symptoms (paresis of the abducens nerve, bulbar disorders, hemiparesis). Further progression of the dissection led to the formation of a dissection aneurysm with a very thin wall, the rupture of which led to massive SAH and subsequent death of the patient.

According to the classification of Yonekura (2002), the formation and growth of aneurysms can occur in four types: type 1 - aneurysm ruptures within a short period of time after formation (days-weeks), type 2 – aneurysm grows slowly (several years) and ruptures when it reaches a certain size, type 3 – aneurysm grows slowly (several years) without rupture. type 4 – the aneurysm grows to a certain size and then remains stable[14]. In this clinical case, a type 1 aneurysm formation was seen. At the same time, when the formation of an aneurysm occurs within a few days or weeks, its pathogenesis is most often associated with inflammation and dilation of the cerebral vessel wall, or with dissection of the wall. Such aneurysms are known as "infectious aneurysms" and "dissection aneurysms". Infectious aneurysms occur in 2-4% of all cerebral aneurysms[15,16], and dissection aneurysms occur in approximately 1% of cases[17].

Infectious aneurysms can form within a few days to several months and have a high risk of rupture[18]. According to John et al. (2016) the risk of aneurysm formation occurs 24 hours after the development of inflammation in the artery wall[19].

The pathogenesis of infectious aneurysms is quite well studied. An inflammation of the cerebral artery wall impairs its ability to resist arterial pressure, leading to over-stretching and aneurysm formation. In most cases, such aneurysms do not have a neck and are fusiform and located in the distal, often cortical branches, of the cerebral arteries. There is little evidence that the formation of infectious aneurysms is also possible in the initial segments of the circle of Willis[15]. In 70-80% of cases, the formation of infectious aneurysms is caused by a bacterial lesion of the artery wall[15,16]. However, there is evidence that they may be based on a viral lesion of the vessel[13].

Keller et al. (2020) performed magnetic resonance imaging (MRI) with a paramagnet in patients with cerebrovascular complications of SARS-CoV-2. In some cases, an accumulation in the walls of large cerebral arteries was detected, indicating an inflammation[20].

According to Castillo et al. (2020) the SARS-CoV-2 virus affects ACE2 receptors that are expressed in the endothelium and muscle cells of intracranial vessels, leading to wall damage and triggering the process of its dissection[10]. A number of researchers suggest such a pathogenesis of vascular dissection against the background of a new coronavirus infection[1,7,13].

In 2020-2021, multiple series of SARS-CoV-2- patients with dissections of extra and intracranial arteries were published[21-23]. Most often dissections of the vessels of the neck and intracranial vessels lead to ischemic disorders of cerebral circulation, but hemorrhagic complications were also reported.

According to Chen et al. (2005), vessel wall dissection between the intima and the muscular layer is most often accompanied by the development of ischemic complications, whereas a dissection between the muscular layer and adventitia is accompanied by the development of aneurysms and hemorrhagic complications[24].

Al-Mufti et al. (2021) studied cerebrovascular complications in patients with SARS-CoV-2, and described three cases of dissection of extra- and intracranial vessels, while in one patient, a dissection of the middle cerebral artery led to massive subarachnoid hemorrhage[1].

There is a pathogenetic relationship between inflammation of the cerebral artery wall, the development of its dissection, and the formation of an aneurysm. Zhong and colleagues (2017) presented a clinical case of a 17-year-old patient who was treated for acute infectious endocarditis. On the tenth day of treatment, the patient developed left-sided ptosis. Additional examinations were performed (MRI and CT angiography, DSA), and a dissection aneurysm of the cavernous part of the internal carotid artery was detected. The patient underwent endovascular embolization of the aneurysm with micro-coils and stent assistance. The operation was uneventful, the ptosis regressed, and the patient was discharged in a good clinical condition[25].

Only isolated cases of cerebral dissection aneurysms in SARS-CoV-2- patients have been described[6,7,13,26].

Savić et al. (2020) published a case of formation and rupture of an aneurysm of the middle cerebral artery in a 13-year-old girl with SARS-CoV-2. The authors believe that the dissection of the wall and the formation of an aneurysm of the M2 segment of the left middle cerebral artery was associated with a viral lesion of the artery. However, they could not completely exclude the possibility of an aneurysm before the disease, which could only provoke its rupture[13].

Al Saiegh et al. (2020) described a case of ruptured dissection aneurysm of the posterior inferior cerebellar artery in a 31-year-old patient with SARS-CoV-2. The patient had a flow-diverter stent placed and the aneurysm was excluded from the blood circulation[26].

In a series of 22 cases of SARS-CoV-2-related cerebrovascular complications, Sweid et al. (2020) noted three cases of ruptured

intracerebral aneurysms. According to the description and presented images, two aneurysms were typical saccular ones, located in the region of branching (in the origin of the posterior communicating artery and in the origin of the anterior choroidal artery), and the aneurysm of the posterior inferior cerebellar artery was dissection type[6].

Dodd et al. (2021) analyzed ruptured cerebral aneurysms in 10 patients with SARS-CoV-2. In 4 cases, aneurysms were dissection type – and three were in the vertebrobasilar territory – which is much more common than among patients with ruptured cerebral aneurysms without coronavirus infection (about 1%). In addition, it was noted that these were younger patients (mean age 38.5 years) than in the general population of patients with SAH (most often in range of 50-60 years)[7].

In all of these studies, there were patients with both severe, mild and even asymptomatic SARS-CoV-2 infections.

Based on all currently available literature, we present the first CT angiography documented case of acute formation of the dissection aneurysm in a patient with SARS-CoV-2.

Conclusion

SARS-CoV-2 appears to have had an impact both on the increase in the frequency of cerebrovascular disorders and on the severity and atypical nature of their clinical course.

A rare, but important, complication of SARS-CoV-2 is a viral lesion of the cerebral artery wall causing its destruction and the formation of a dissection aneurysm with a very thin wall and a high risk of rupture.

In patients with SARS-CoV-2 and non-traumatic SAH and ICH, it is necessary to carefully examine the condition of cerebral vessels for dissections and dissection aneurysms. In unclear cases, as well as with small aneurysms, cerebral angiography has maximum informative value.

In patients with SARS-CoV-2, the first line of treatment for dissection aneurysms (with or without rupture) may be endovascular exclusion (embolization with stent assistance, placement of flow diverter stents). When endovascular exclusion of a dissection aneurysm is not possible, it may be necessary to consider possible options for microsurgical treatment.

Disclosures

Conflict of Interest: All authors certify that they have no affiliations with or involvement in any organization or entity with any financial interest (such as honoraria; educational grants; participation in speakers' bureaus; membership, employment, consultancies, stock ownership, or other equity interest; and expert testimony or patent-licensing arrangements), or non-financial interest (such as personal or professional relationships, affiliations, knowledge or beliefs) in the subject matter or materials discussed in this manuscript.

Ethical approval: All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee (Krasnoyarsk State Medical University named after Prof. V.F. Voino-Yasenetsky and FSBI "Federal Center of Neurosurgery", Novosibirsk) and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

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References

1. Al-Mufti F, Becker C, Kamal H, et al. Acute Cerebrovascular Disorders and Vasculopathies Associated with Significant Mortality in SARS-CoV-2 Patients Admitted to The Intensive Care Unit in The New York Epicenter. J Stroke Cerebrovasc Dis. 2021;30(2):105429. doi: 10.1016/j.jstrokecerebrovasdis.2020.105429..
2. Carod-Artal FJ. Neurological complications of coronavirus and COVID-19. Rev Neurol. 2020;70(9):311-322. doi: 10.33588/rn.7009.2020179.
3. Katsanos AH, Palaiodimou L, Zand R, et al. The Impact of SARS-CoV-2 on Stroke Epidemiology and Care: A Meta-Analysis. Ann Neurol. 2021;89(2):380-388. doi: 10.1002/ana.25967.
4. Maury A, Lyoubi A, Peiffer-Smadja N, de Broucker T, Meppiel E. Neurological manifestations associated with SARS-CoV-2 and other coronaviruses: A narrative review for clinicians. Rev Neurol (Paris). 2021;177(1-2):51-64. doi: 10.1016/j.neurol.2020.10.001.
5. Ravindra VM, Grandhi R, Delic A, et al. Impact of COVID-19 on the hospitalization, treatment, and outcomes of intracerebral and subarachnoid hemorrhage in the United States. PLoS One. 2021;16(4):e0248728. Published 2021 Apr 14. doi: 10.1371/journal.pone.0248728
6. Sweid A, Hammoud B, Bekelis K, et al. Cerebral ischemic and hemorrhagic complications of coronavirus disease 2019. Int J Stroke. 2020;15(7):733-742. doi: 10.1177/1747493020937189.

7. Dodd WS, Jabbour PM, Sweid A, et al. Aneurysmal Subarachnoid Hemorrhage in Patients with Coronavirus Disease 2019 (COVID-19): A Case Series. World Neurosurg. 2021;153:e259-e264. doi: 10.1016/j.wneu.2021.06.092
8. Nawabi J, Morotti A, Wildgruber M, et al. Clinical and Imaging Characteristics in Patients with SARS-CoV-2 Infection and Acute Intracranial Hemorrhage. J Clin Med. 2020;9(8):2543. Published 2020 Aug 6. doi: 10.3390/jcm9082543
9. Zhou Z, Kang H, Li S, Zhao X. Understanding the neurotropic characteristics of SARS-CoV-2: from neurological manifestations of COVID-19 to potential neurotropic mechanisms. J Neurol. 2020;267(8):2179-2184. doi: 10.1007/s00415-020-09929-7.
10. Castillo PR, Del Brutto OH, Mautong H, et al. Hemorrhagic stroke in hispanics with severe SARS-CoV2 infection. J Neurol Sci. 2020;416:117046. doi: 10.1016/j.jns.2020.117046
11. Qureshi AI, Baskett WI, Huang W, et al. Subarachnoid Hemorrhage and COVID-19: An Analysis of 282,718 Patients. World Neurosurg. 2021;151:e615-e620. doi: 10.1016/j.wneu.2021.04.089
12. Rustemi O, Raneri F, Iannucci G, Volpin L, Segna A. Aneurysmal subarachnoid hemorrhage in a SARS-CoV-2 positive testing: casual or causal?. Br J Neurosurg. 2022;36(2):293-294. doi: 10.1080/02688697.2020.1787343.
13. Savić D, Alsheikh TM, Alhaj AK, et al. Ruptured cerebral pseudoaneurysm in an adolescent as an early onset of COVID-19 infection: case report. Acta Neurochir (Wien). 2020;162(11):2725-2729. doi: 10.1007/s00701-020-04510-7.
14. Yonekura M. Importance of prospective studies for deciding on a therapeutic guideline for unruptured cerebral aneurysm. Acta Neurochir Suppl. 2002;82:21-25. doi: 10.1007/978-3-7091-6736-6_4
15. Alawieh A, Chaudry MI, Turner RD, Turk AS, Spiotta AM. Infectious intracranial aneurysms: a systematic review of epidemiology, management, and outcomes. J Neurointerv Surg. 2018;10(7):708-716. doi: 10.1136/neurintsurg-2017-013603
16. Ducruet AF, Hickman ZL, Zacharia BE, et al. Intracranial infectious aneurysms: a comprehensive review. Neurosurg Rev. 2010;33(1):37-46. doi: 10.1007/s10143-009-0233-1
17. Daou B, Hammer C, Chalouhi N, et al. Dissecting pseudoaneurysms: predictors of symptom occurrence, enlargement, clinical outcome, and treatment. J Neurosurg. 2016;125(4):936-942. doi: 10.3171/2015.10.JNS151846
18. Kannoth S, Iyer R, Thomas SV, et al. Intracranial infectious aneurysm: presentation, management and outcome. J Neurol Sci. 2007;256(1-2):3-9. doi: 10.1016/j.jns.2007.01.044.
19. John S, Walsh KM, Hui FK, Sundararajan S, Silverman S, Bain M. Dynamic Angiographic Nature of Cerebral Mycotic Aneurysms in Patients With Infective Endocarditis. Stroke. 2016;47(1):e8-e10. doi: 10.1161/STROKEAHA.115.011198.
20. Keller E, Brandi G, Winklhofer S, et al. Large and Small Cerebral Vessel Involvement in Severe COVID-19: Detailed Clinical Workup of a Case Series. Stroke. 2020;51(12):3719-3722. doi: 10.1161/STROKEAHA.120.031224
21. Morassi M, Bigni B, Cobelli M, Giudice L, Bnà C, Vogrig A. Bilateral carotid artery dissection in a SARS-CoV-2 infected patient: causality or coincidence?. J Neurol. 2020;267(10):2812-2814. doi: 10.1007/s00415-020-09984-0.
22. Patel P, Khandelwal P, Gupta G, Singla A. "COVID-19 and cervical artery dissection- A causative association?". J Stroke Cerebrovasc Dis. 2020;29(10):105047. doi: 10.1016/j.jstrokecerebrovasdis.2020.105047
23. Salik I, Jacoby M. Carotid Artery Dissection and Hemorrhagic Stroke in the Setting of Multisystem Inflammatory Syndrome in Children. Cureus. 2021;13(3):e13640. Published 2021 Mar 1. doi: 10.7759/cureus.13640.
24. Chen M, Caplan L. Intracranial dissections. Front Neurol Neurosci. 2005;20:160-173. doi: 10.1159/000088166
25. Zhong X, Li X, Shao S, Yang X, Fan X. A case of infectious intracranial dissecting aneurysm. Neurol India. 2017;65(2):405-407. doi: 10.4103/neuroindia.NI_1223_15.
26. Al Saiegh F, Ghosh R, Leibold A, et al. Status of SARS-CoV-2 in cerebrospinal fluid of patients with COVID-19 and stroke. J Neurol Neurosurg Psychiatry. 2020;91(8):846-848. doi: 10.1136/jnnp-2020-323522.

CASE REPORT

Primary pituitary lymphoma successfully treated with Bruton's tyrosine kinase inhibitor monotherapy: case report

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Abstract

Introduction: Primary pituitary lymphoma (PPL) is a rare disease characterized by lymphoma confined to the sella or parasellar region without systemic involvement. The clinical symptoms of PPL may include headache, hypopituitarism, visual field disturbance and visual impairment. To date, there is no established standard treatment for this condition.

Here, we present a case of successful treatment with a Bruton's tyrosine kinase (BTK) inhibitor.

Case report: A 78-year-old man with a history of severe left renal insufficiency caused by retroperitoneal fibrosis, and sequential right nephrostomy, underwent brain magnetic resonance imaging (MRI) due to the altered hormonal status. An enlarged pituitary stalk was noted and led to a diagnosis of lymphocytic hypopituitarism.

Six months later, visual field disturbance and visual acuity deterioration developed, and an MRI revealed a neoplastic lesion and further enlargement of the stalk and the pituitary itself, with an obvious optic nerve compression. Expedited transsphenoidal partial resection was performed to relieve the compression. Pathophysiology led to the diagnosis of the large B-cell lymphoma of the germinal center origin. Because of the patient's poor renal function, high-dose methotrexate therapy was not an option; rather, the patient was treated with a BTK inhibitor - tirabrutinib. Symptoms improved within a week, and a follow-up MRI confirmed a marked reduction of the pituitary lesion.

Conclusion: BTK inhibitors may be considered as a first-line treatment option for PPL, especially in patients with contraindications for other treatment protocols.

Keywords: hypopituitarism; malignant lymphoma; primary pituitary lymphoma; Bruton's tyrosine kinase inhibitor; tirabrutinib

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Introduction

Malignant lymphomas originating from the central nervous system, or primary central nervous system lymphoma (PCNSL) account for about 3% of intracranial malignancies[1], with those arising within the sella turcica region being extremely rare[2]. Primary malignant lymphoma of the pituitary gland (primary pituitary lymphoma, PPL) is generally considered when a lymphoma is confined to the sella or parasellar region, without systemic invasion[3]. The relevant clinical symptoms of include headache, hypopituitarism, and visual field and acuity disturbances.

Primary pituitary lymphoma is extremely rare, lacks evidence regarding treatment, and is typically treated the same way as PCNSL[4]. High-dose methotrexate (MTX) and the whole-brain radiation therapy of 30–40 Gy, is the standard of treatment for newly diagnosed PCNSL in Japan. In patients with relapsed disease, high-dose MTX with rituximab-based retreatment; monotherapy with rituximab, topotecan, and temsirolimus; as well as the Bruton's tyrosine kinase (BTK) inhibitors; have been tried, but the optimal therapy is yet to be elucidated[5].

Bruton's tyrosine kinase (BTK), a downstream mediator of the B-cell antigen receptor (BCR), has been linked to a number of B-cell malignancies. BTK inhibitors suppress tumor cell growth by inhibiting BTK activity, and have been shown to be effective against PCNSL[5]. Tirabrutinib is a second-generation BTK inhibitor that is more potent and selective than those of the first generation[6]. It is approved in Japan for the treatment of relapsed/refractory PCNSL, and it's urine excretion accounts for only about 10% of the total[5].

With a PPL only partially resected, and limited chemotherapeutic options due to the renal insufficiency, we aimed to complement the initial surgery with tirabrutinib monotherapy.

Case report

A 45-year-old male patient was admitted to the infectious diseases hospital on December 18, 2021. He was treated for a SARS-CoV-2 infection complicated by bilateral poly-segmental pneumonia. Since admission, the patient had a severe headache (up to 8 points on a visual analog scale).

A 78-year-old male was referred to our department by an ophthalmologist because of visual field disturbance and visual acuity deterioration; with a suspected pituitary neoplasm. Except for the bitemporal hemianopsia, the patients other neurological status was unremarkable.

Six months prior, the patient was diagnosed with lymphocytic hypopituitarism after brain magnetic resonance imaging (MRI) revealed an enlarged pituitary stalk. Anterior and posterior pituitary hormones secretions were insufficient, and hormone replacement therapy involving levothyroxine, hydrocortisone, and desmopressin was started. Symptoms improved, and he was followed up with outpatient visits.

Repeated MRI revealed a pituitary lesion, enlarging the pituitary gland and the stalk, extending from the sella turcica to the cephalic region and compressing the optic nerve (***Figure 1***).

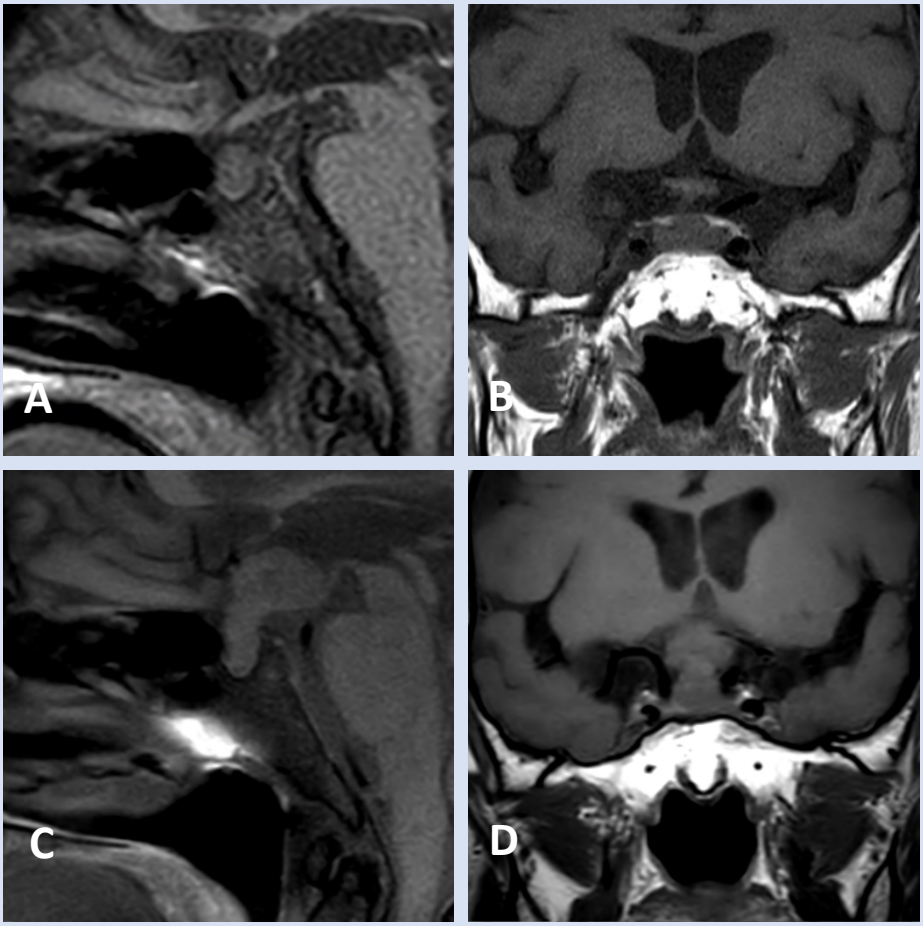

Figure 1. MRI simple T1-weighted image (sagittal and coronal planes): A. at the first visit to the previous hospital; B. the tumor had enlarged 6 months after the first visit.

The patient had a medical history of retroperitoneal fibrosis, left renal insufficiency, and right nephrostomy, which required regular replacement. Blood laboratory tests revealed low levels of total protein, albumin, and an estimated glomerular filtration rate (32.6 mL/min/L), and elevated levels of C-reactive protein, IgG, IgA, IgM, IgG4, prolactin, and anti-neutrophil cytoplasmic antibody. The patient also exhibited low levels of anterior pituitary hormones and slightly elevated levels of ProGRP (90.1 g/mL) and soluble IL-2 receptor (661 U/mL); however, the tumor markers were negative. IgG4-related hypopituitarism was considered, therefore, a whole-body FDG positron emission tomography (PET) scan and CSF examination were performed, but no abnormalities were detected.

Since the visual field disturbances and visual impairment progressed, an expedited transsphenoidal resection was performed.

Histopathological examination revealed a lesion of lymphocyte-like atypical cell proliferation with a necrotic background. Immunohistochemistry indicated that the atypical cells were positive for CD20 and BCL6 staining and negative for MUM1 staining, and had an MIB1 labeling index of ~70%; thus, a diagnosis of large B-cell lymphoma of germinal center origin was established (***Figure 2***). Bone marrow aspiration, to investigate the possibility of bone marrow involvement, obtained no malignant cells.

Although high-dose MTX is the recommended first-line treatment according to the national protocol for PCNSL[7], because of his renal impairment and age, the patient received oral tirabrutinib. Within a week of starting oral administration of this drug, the lesion exhibited shrinkage, and the patient's visual field disturbance and visual acuity improved. However, after 6 weeks of oral administration, severe pneumonia caused by cytomegalovirus emerged, leading to the discontinuation of tirabrutinib. Eight weeks after discontinuing the treatment, a follow-up MRI showed that the tumor disappearance was maintained (***Figure 3***).

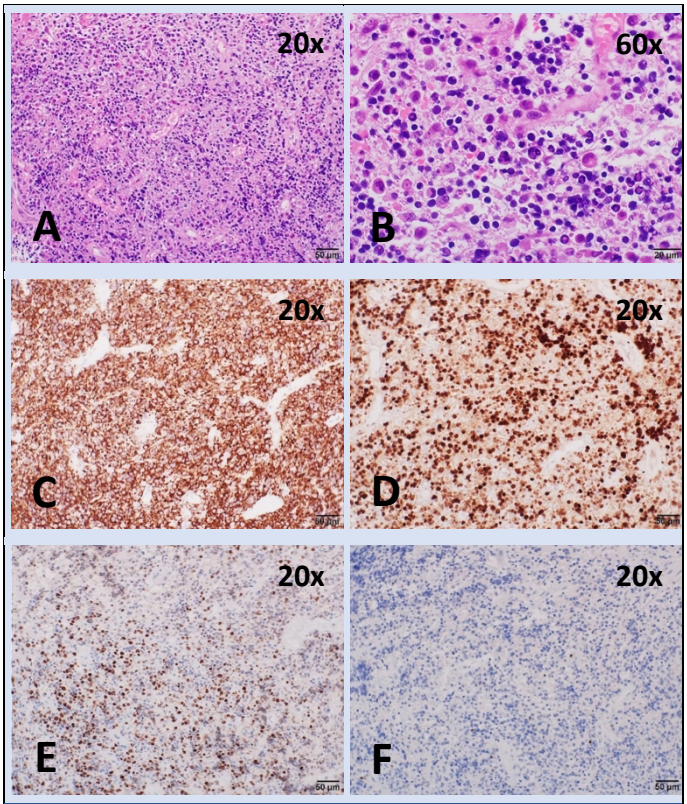

Figure 1. Pathology of the pituitary tumor biopsy. A. 20x hematoxylin-eosin staining showed a lesion consisting of lymphocyte-like atypical cell proliferation on a background of necrosis and immunostaining B. 60x: C. CD20 staining positive, D. MIB1 labeling rate about 70%, E. BCL6 staining positive, and F. MUM1 staining negative led to a diagnosis of large B-cell lymphoma, germinal center B-cell-like type.

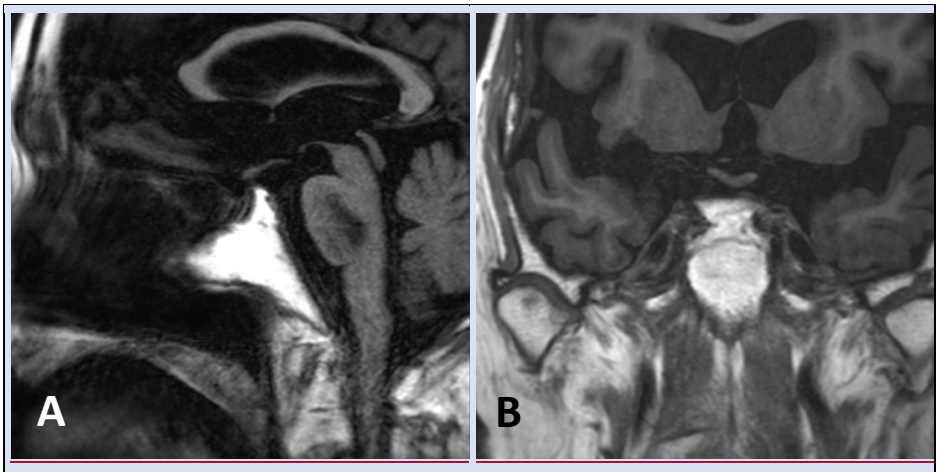

Figure 1. MRI T1-weighted image (sagittal and coronal planes) 8 weeks after chemotherapy.

Discussion

To the best of our knowledge, this is the first case report of PPL treated with tirabrutinib. Even more, the treatment was successful, and the BTK inhibitor was used as an adjuvant monotherapy after partial surgical resection.

Primary central nervous system lymphoma involving only the pituitary gland is rare, with only about 40 adult cases reported to time. Due to its specifies, one previous study suggested that PPL should be considered a separate entity from PCNSL, as the pituitary gland (as well as the pineal) lack a blood–brain barrier and pose a different embryological structure from the brain parenchyma, thus constituting for distinguish[4,8].

Diffuse large B-cell lymphoma is the most common type of PCNSL[9] and this applies to the PPL as well. With the emerge of the gene expression profiling, two subgroups were identified: the germinal center B cell-like (GCB); and the activated B cell-like and heterogenous subtypes (Non-GCB)[10]. Unlike our case, the activated B cell-like is the most common subtype of PCNSL, usually ascribed as the reason for the poor prognosis[11]. BTK inhibitors inhibit the proliferation and induce apoptosis of GCB PNCSL cell lines, and appear efficient in this particular subtype[12], thus leading to the good response in our patient as well.

Primary central nervous system lymphoma is generally treated with chemotherapy based on high-dose MTX, which bypasses the blood–brain barrier, but since the pituitary gland does not have a blood–brain barrier it allows for the wider variety of efficient options including: high-dose MTX[13]; cyclophosphamide, doxorubicin hydrochloride, vincristine sulfate, and prednisone (CHOP) therapy[14]; but also, other protocols with varying efficacy[3,15,16]. Because of impaired renal function and the patients advanced age, neither high-dose MTX-based nor CHOP therapy[7] were considered an option in our case, and oral tirabrutinib was chosen instead.

Surgery is usually not considered in PCNSL[17], however, due to the frequent compression-related symptoms in PPL, partial resection to debulk the lesion and obtain histopathology was fully justified, especially due to the worsening despite symptomatic treatment. Radiotherapy was frequently used in PPL[18,19], but, due to the increased toxicity[20], recently reported therapies, omitted the radiotherapy from their armamentarium[3,4,21], and so did us.

The tumor exhibited marked shrinkage 1 week after the tirabrutinib administration was started. Regardless of the premature treatment discontinuation after 6 weeks because of severe infection and omitted radiotherapy, complete remission was maintained for the next 2 months, suggesting that the treatment was ultimately safe and effective.

Conclusion

After initial surgical resection and histopathological confirmation, tirabrutinib appears as a viable alternative treatment option for patients with PPL, even more so in patients who are unable to receive other agents for any given reason. Future prospective studies are needed to establish the efficacy of BTK inhibitor chemotherapy on the outcome and prognosis of patients with PPL.

Disclosures

Conflict of Interest: All authors certify that they have no affiliations with or involvement in any organization or entity with any financial interest (such as honoraria; educational grants; participation in speakers' bureaus; membership, employment, consultancies, stock ownership, or other equity interest; and expert testimony or patent-licensing arrangements), or non-financial interest (such as personal or professional relationships, affiliations, knowledge or beliefs) in the subject matter or materials discussed in this manuscript.

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References

1. Giustina A, Gola M, Doga M, Rosei EA. Clinical review 136: Primary lymphoma of the pituitary: an emerging clinical entity. J Clin Endocrinol Metab. 2001;86(10):4567-4575. doi:10.1210/jcem.86.10.7909
2. Freda PU, Post KD. Differential diagnosis of sellar masses. Endocrinol Metab Clin North Am. 1999;28(1):81-vi. doi:10.1016/s0889-8529(05)70058-x
3. Duan L, Liu J, Zhang Y, et al. Primary Pituitary Lymphoma in Immunocompetent Patients: A Report on Two Case Studies and the Review of Literature. Front Endocrinol (Lausanne). 2021;11:562850. Published 2021 Feb 4. doi:10.3389/fendo.2020.562850
4. Tarabay A, Cossu G, Berhouma M, Levivier M, Daniel RT, Messerer M. Primary pituitary lymphoma: an update of the literature. J Neurooncol. 2016;130(3):383-395. doi:10.1007/s11060-016-2249-z
5. Narita Y, Nagane M, Mishima K, et al. Phase I/II study of tirabrutinib, a second-generation Bruton's tyrosine kinase inhibitor, in relapsed/refractory primary central nervous system lymphoma. Neuro Oncol. 2021;23(1):122-133. doi:10.1093/neuonc/noaa145
6. Okita Y, Kano-Fujiwara R, Nakatsuka SI, Honma K, Kinoshita M. Histological verification of the treatment effect of tirabrutinib for relapsed/refractory primary central nervous system lymphoma. Exp Hematol Oncol. 2021;10(1):29. doi:10.1186/s40164-021-00222-5
7. Nakamura H, Takami H, Yanagisawa T, et al. The Japan Society for Neuro-Oncology guideline on the diagnosis and treatment of central nervous system germ cell tumors. Neuro Oncol. 2022;24(4):503-515. doi:10.1093/neuonc/noab242
8. Konishi J, Miki Y, Asato R. Imaging Diagnosis of Hypothalamic Pituitary Diseases. Nihon Naika Gakkai Zasshi. 1994;83(12):2117-21. doi: 10.2169/naika.83.2117.
9. Deckert M, Engert A, Brück W, et al. Modern concepts in the biology, diagnosis, differential diagnosis and treatment of primary central nervous system lymphoma. Leukemia. 2011;25(12):1797-1807. doi:10.1038/leu.2011.169
10. Mahadevan A, Rao CR, Shanmugham M, Shankar SK. Primary central nervous system diffuse large B-cell lymphoma in the immunocompetent: Immunophenotypic subtypes and Epstein-Barr virus association. J Neurosci Rural Pract. 2015;6(1):8-14. doi:10.4103/0976-3147.143171
11. Yang XL, Liu YB. Advances in Pathobiology of Primary Central Nervous System Lymphoma. Chin Med J (Engl). 2017;130(16):1973-1979. doi:10.4103/0366-6999.211879
12. Zheng X, Ding N, Song Y, Feng L, Zhu J. Different sensitivity of germinal center B cell-like diffuse large B cell lymphoma cells towards ibrutinib treatment. Cancer Cell Int. 2014;14(1):32. Published 2014 Apr 3. doi:10.1186/1475-2867-14-32
13. Quintero Wolfe S, Hood B, Barker J, Benveniste RJ. Primary central nervous system lymphoma mimicking pituitary apoplexy: case report. Pituitary. 2009;12(1):76-79. doi:10.1007/s11102-008-0084-8
14. Rudnik A, Larysz D, Blamek S, et al. Primary pituitary lymphoma. Folia Neuropathol. 2007;45(3):144-148.
15. Ogawa A, Sugano H, Inoue T, Fujihara M, Matsumoto S, Tsuchiyama Y, et al. Malignant Lymphoma Presented with Thickned Pituitary Stalk and Hypopituitalism. Nihon Naika Gakkai Zasshi. 2008;97(6):1324-6. doi: 10.2169/naika.97.1324.
16. Huang YY, Lin SF, Dunn P, Wai YY, Hsueh C, Tsai JS. Primary pituitary lymphoma presenting as hypophysitis. Endocr J. 2005;52(5):543-549. doi:10.1507/endocrj.52.543
17. Schaff LR, Grommes C. Primary central nervous system lymphoma. Blood. 2022;140(9):971-979. doi:10.1182/blood.2020008377
18. Ravindra VM, Raheja A, Corn H, et al. Primary pituitary diffuse large B-cell lymphoma with somatotroph hyperplasia and acromegaly: case report. J Neurosurg. 2017;126(5):1725-1730. doi:10.3171/2016.5.JNS16828
19. Romeike BF, Joellenbeck B, Stein H, et al. Precursor T-lymphoblastic lymphoma within a recurrent pituitary adenoma. Acta Neurochir (Wien). 2008;150(8):833-836. doi:10.1007/s00701-008-1585-y
20. Thiel E, Korfel A, Martus P, et al. High-dose methotrexate with or without whole brain radiotherapy for primary CNS lymphoma (G-PCNSL-SG-1): a phase 3, randomised, non-inferiority trial. Lancet Oncol. 2010;11(11):1036-1047. doi:10.1016/S1470-2045(10)70229-1
21. Ban VS, Chaudhary BR, Allinson K, Santarius T, Kirollos RW. Concomitant Primary CNS Lymphoma and FSH-Pituitary Adenoma Arising Within the Sella. Entirely Coincidental? Neurosurgery. 2017;80(1):E170-E175. doi: 10.1093/neuros/nyw003

NEUROSONOLOGY X NEUROSURGERY

Insonation angle impact on micro-Doppler evaluation in cerebrovascular surgery: technical note

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Abstract

Introduction: The insonation angle consideration is important in the Doppler-sonographic evaluation of blood flow velocities and volume, however, the concept is rarely applied to the intraoperatively used micro-Doppler.

This technical note aims to emphasize the possibility and necessity of insonation angle correction and preservation for the blood flow assessment in cerebrovascular surgery.

Methods: Bi-directional surgical Doppler with a 20MHz flexible bayonet 1.2mm diameter probe was used for the intraoperative blood flow assessment, with the variable insonation angle on the straight parts of the arteries, and the results were recorded to demonstrate the importance.

Results: The measurements performed confirmed that the blood flow features, including the direction, velocity, and especially the volume, are highly susceptible to the insonation angle variations. Although there were significant alterations, positioning the probe at an angle of ~60 degrees resulted in the variable but less altering measurements.

Conclusion: Positioning the probe at ~60 degrees is desirable; however, it appears that it is even more important to maintain the same insonation angle and probe position when assessing the vessels repeatedly for blood flow alterations that might occur in the course of cerebrovascular surgery.

Keywords: micro-Doppler; insonation angle; cerebrovascular surgery

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Introduction

Intraoperative micro-Doppler blood flow evaluation is one of the most useful tools in cerebrovascular surgery[1], allowing to recognize inadvertent vessel narrowing, or incomplete aneurysm occlusion[2,3], bypass effects and patency[4], and perform stepwise AVM elimination[5]. In order to obtain relevant measurement results, at least the basics of micro-Doppler use should be mastered.

The insonation angle consideration is indispensable in evaluation of normal values, physiological variations, as well as in the evaluation of pathological findings of blood flow velocities in Doppler examination[6,7]. Inappropriate Doppler-sonographic measurement may lead to the false satisfactory results, due to the insonation angle differences between examinations, leading to the oversight of the blood flow disturbance, or deeming the altered blood flow appropriate[8].

Transcranial color-coded duplex sonography (TCCD) introduced simultaneous visualization of the vessels in color, together with Doppler measurement of blood flow velocities and made the correction of the insonation angle possible, resulting in more realistic values of flow velocities for intracranial arteries. After several papers emphasized the importance of the insonation angle[6,8,9], the function to adjust insonation angle was added to almost every ultrasound system.

On the other hand, not much attention was drawn to the concept in the intraoperative use of micro-Doppler. With a clear view of the vessels to be examined it was rarely considered in the literature, while the devices used do not allow for the adjustment of the insonation angle by software means[10]. In the recent study, neuronavigation was used to perform the insonation at an appropriate insonation angle[11].

This technical note aims to emphasize the possibility and necessity of insonation angle correction during the intraoperative cerebral blood vessel assessment, without any additional burden.

Methods

Micro-Doppler sonography is routinely used in our department for all cerebrovascular cases undergoing surgery for aneurysm, arteriovenous malformation or dural arteriovenous fistula, as well as those receiving a bypass surgery for any underlying pathology[5,12].

Bi-directional surgical Doppler DVM 4300 with a 20MHz flexible bayonet 1.2mm diameter probe (Hadeco, Kawasaki, Japan) was used for the intraoperative blood flow assessment, while watery fluid environment was provided by the probe and vessels irrigation flushing.

Since the probe placement at the position for the measurement of maximum (real) velocity is usually impossible (0 degrees in relation to the blood flow direction), in clinical settings, it is usually recommended to strive towards ~60 degrees angle of insonation[13].

For the purpose of this technical note, the blood flow was evaluated with the variable insonation angle varying from 30 to 150 degrees to the blood flow direction, on the straight parts of the arteries and the results were recorded to demonstrate the importance, while the velocity of ~0, notable at 90 degrees was used to emphasize this feature[14]. ***(Figure 1)***.

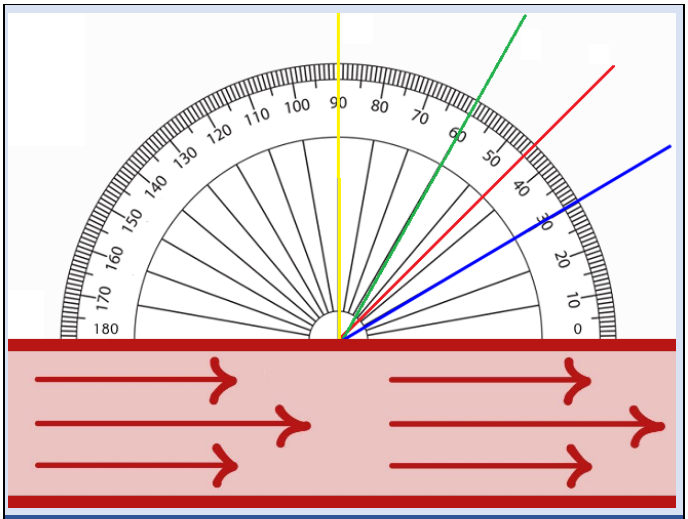

Figure 1. Various positionings of the micro-Doppler probe to the blood flow within the vessel

Results

The measurements performed confirmed that the blood flow features, including the direction velocity, and especially the volume is highly susceptible to the insonation angle variations.

The velocity direction varied when the probe was aligned at an angle towards the flow, and when aligned opposite to the expected flow direction (<90 and >90 degrees respectively).

Minimal blood flow velocity values (+5 to –5 cm/s) were measured when the probe was placed at ~90 degrees to the vessel flow, while the velocities of up to 50cm/s were measured when the probe was placed at 45-60 degrees. These differences appear even more accentuated when considering the volumes with variations ranging from 10 to more than 200 ml/s with these same insonation angle placements (***Figure 2***).

Although there were significant alterations, in consistency with the previous study[14], positioning the probe at an angle anywhere between 45-60 degrees resulted in the variable but less altering measurements.

Discussion

Digital subtractional angiography is a gold standard in the blood flow evaluation in cerebrovascular surgery, however, it is ergonomically and economically disadvantaged, and time-consuming. Indocyanine green (ICG) videoangiography appears as a rightfull successor, but ICG-videoangiography is not able to replace micro-Doppler, as it cannot determine the velocity characteristics of the blood flow. Moreover, micro-Doppler sonography remains frequently used mainly due to the ease of use, repeatability, and time efficiency[15].

The velocity, measured by Doppler ultrasound systems, is physically dependent on the cosine function of the angle between the blood flow and the direction of the ultrasound emitted by the probe. The cosine of a 90-degree angle is zero, therefore, measuring velocities at an angle close to 90 degrees gives falsely low values of blood flow velocities[16].

With the introduction of Duplex sonography, which implies the simultaneous application of imaging (2D or B-mode) simultaneously with Doppler, and especially after the introduction of color Doppler (Triplex), the possibility of correcting the insonation angle based on the imaging of the blood vessel and the direction of the Doppler beam was obtained. With the feature introduction, awareness was raised, and nowadays, angle of insonation implementation is considered a common practice in transcranial Doppler-sonography[17].

Although intraoperative use allows for the direct visual control of the angle of insonation, there are a few features, which usually limit the micro-Doppler probe manipulation, namely the constrained operative field (limited by the size of craniotomy, extent of Sylvian fissure dissection, brain edema) and the microscope use, with altered visualization planes, leading to the inaccuracies when considering a certain angle is achieved[18] (***Figure 2***).

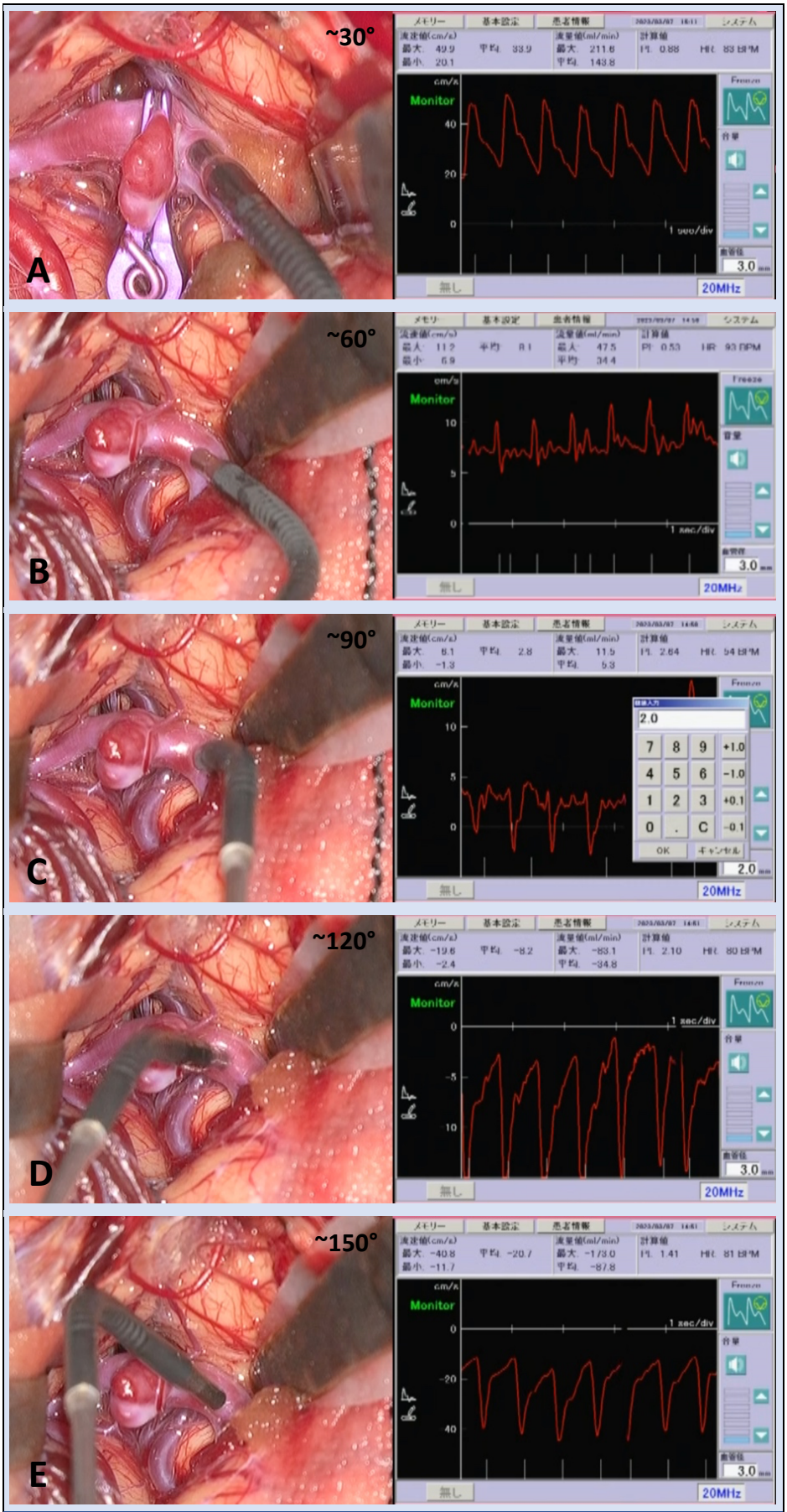

Figure 2. Various angles of insonation used for the same artery assessment showing significant variability of blood flow velocities and volumes, at the: A. ~30; B. ~60; C. ~90; D. 120; and E. 150; degrees insonation angles. Note the inaccuracy in angle perception.

To overcome these the limitations related to the operative field, as the features should not be corrected purely to accommodate micro-Doppler probe, achieving the desired angle may turn out to be impossible, it is important to maintain a same (or similar, at least) angle in the two measurements. In our experience, it is usually useful to lean the probe against the brain parenchyma, dura, or bony surface while keeping the same spot on the vessel when replacing the probe. The accuracy of replacement could be further improved with dye markings on the vessel wall.

As it was previously reported, the variations of the blood flow velocity and volume vary significantly between 0-90 degrees, however, for the same inaccuracies of 10 degrees, the differences are less pronounced when the desired angle is about 60 degrees, when compared to 80 or 30[14]. Another possibility is use of the negative (mirror) flow measurement, when the probe is placed >90 degrees, to achieve better positioning and more favorable angle by the utilization of the wider space on the contralateral side.

Finally, to overcome the microscope related inaccuracy, it is always possible to position the probe under naked-eye, however, this manipulation might be dangerous, and it could get complicated in those without 20/20 vision, when using microscope diopter adjustment feature for vision correction. Nevertheless, when used in an exoscopic environment, the chances of an error diminish.

Conclusion

Based on the previous recommendations and our experience it is usually most appropriate to position the probe at ~60 degrees, however, it appears that it is even more important to maintain the same insonation angle and probe position when assessing the vessels repeatedly to capture the valid blood flow alterations that might occur in the course of cerebrovascular surgery.

Disclosures

Conflict of Interest: All authors certify that they have no affiliations with or involvement in any organization or entity with any financial interest (such as honoraria; educational grants; participation in speakers' bureaus; membership, employment, consultancies, stock ownership, or other equity interest; and expert testimony or patent-licensing arrangements), or non-financial interest (such as personal or professional relationships, affiliations, knowledge or beliefs) in the subject matter or materials discussed in this manuscript.

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References

1. Dubovoy A, Lekchnov E, Galaktionov D, Ovsyannikov K, Bervitskiy A, Sosnov A, et al. Microsurgical management of complex middle cerebral artery aneurysms. Neurohirurgija - The Serbian Journal of Neurosurgery. 2021;1(1). doi: 10.55005/sjns.v1i1.2.
2. Firsching R, Synowitz HJ, Hanebeck J. Practicability of intraoperative microvascular Doppler sonography in aneurysm surgery. Minim Invasive Neurosurg. 2000;43(3):144-8. doi: 10.1055/s-2000-12261.
3. Cui H, Wang Y, Yin Y, Wan J, Fei Z, Gao W, et al. Role of intraoperative microvascular Doppler in the microsurgical management of intracranial aneurysms. J Clin Ultrasound. 2011;39(1):27-31. doi: 10.1002/jcu.20751.
4. Morisawa H, Kawamata T, Kawashima A, Hayashi M, Yamaguchi K, Yoneyama T, et al. Hemodynamics and changes after STA-MCA anastomosis in moyamoya disease and atherosclerotic cerebrovascular disease measured by micro-Doppler ultrasonography. Neurosurg Rev. 2013;36(3):411-9. doi: 10.1007/s10143-012-0441-y.
5. Burkhardt T, Siasios G, Schmidt NO, Reitz M, Regelsberger J, Westphal M. Intraoperative Micro-Doppler in Cerebral Arteriovenous Malformations. J Neurol Surg A Cent Eur Neurosurg. 2015;76(6):451-5. doi: 10.1055/s-0035-1551829.
6. Krejza J, Mariak Z, Babikian VL. Importance of angle correction in the measurement of blood flow velocity with transcranial Doppler sonography. AJNR Am J Neuroradiol. 2001;22(9):1743-7. doi:
7. Lepic T, Veljancic D, Jovanikic O, Lepic M, Slankamenac P, Raicevic R. Importance of angle correction in transcranial color-coded duplex insonation of arteries at the base of the brain. Vojnosanit Pregl. 2015;72(12):1093-7. doi: 10.2298/vsp141105009l.
8. Eicke BM, Tegeler CH, Dalley G, Myers LG. Angle correction in transcranial Doppler sonography. J Neuroimaging. 1994;4(1):29-33. doi: 10.1111/jon19944129.
9. Bartels E, Flugel KA. Quantitative measurements of blood flow velocity in basal cerebral arteries with transcranial duplex color-flow imaging. A comparative study with conventional transcranial Doppler sonography. J Neuroimaging. 1994;4(2):77-81. doi: 10.1111/jon19944277.
10. Lepić M. Intraoperative ultrasound use in cranial neurosurgery: The basics and initial experience. Neurohirurgija - The Serbian Journal of Neurosurgery. 2022;1(1):39-44. doi: 10.55005/sjns.v1i1.4.
11. Malinova V, von Eckardstein K, Rohde V, Mielke D. Neuronavigated microvascular Doppler sonography for intraoperative monitoring of blood flow velocity changes during aneurysm surgery - a feasible monitoring technique. Clin Neurol Neurosurg. 2015;137:79-82. doi: 10.1016/j.clineuro.2015.06.021.
12. Bhave VM, Stone LE, Rennert RC, Steinberg JA. Complementary Tools in Cerebral Bypass Surgery. World Neurosurg. 2022;163:50-9. doi: 10.1016/j.wneu.2022.03.146.
13. Revzin MV, Imanzadeh A, Menias C, Pourjabbar S, Mustafa A, Nezami N, et al. Optimizing Image Quality When Evaluating Blood Flow at Doppler US: A Tutorial. Radiographics. 2019;39(5):1501-23. doi: 10.1148/rg.2019180055.
14. Klünemann HH, Arnolds BJ, von Reutern GM. Angle dependence and transit time effect in Doppler flow measurement. European Journal of Ultrasound. 1996;3(3):261-6. doi: 10.1016/0929-8266(95)00158-n.
15. Nickele C, Nguyen V, Fisher W, Couldwell W, Aboud E, David C, et al. A Pilot Comparison of Multispectral Fluorescence to Indocyanine Green Videoangiography and Other Modalities for Intraoperative Assessment in

Vascular Neurosurgery. Operative Neurosurgery. 2019;17(1):103-9. doi: 10.1093/ons/opy237.
16. Oglat AA, Matjafri MZ, Suardi N, Oqlat MA, Abdelrahman MA, Oqlat AA. A Review of Medical Doppler Ultrasonography of Blood Flow in General and Especially in Common Carotid Artery. J Med Ultrasound. 2018;26(1):3-13. doi: 10.4103/jmu.Jmu_11_17.
17. Alexandrov AV, Sloan MA, Wong LK, Douville C, Razumovsky AY, Koroshetz WJ, et al. Practice standards for transcranial Doppler ultrasound: part I--test performance. J Neuroimaging. 2007;17(1):11-8. doi: 10.1111/j.1552-6569.2006.00088.x.
18. Siasios I, Kapsalaki EZ, Fountas KN. The role of intraoperative micro-Doppler ultrasound in verifying proper clip placement in intracranial aneurysm surgery. Neuroradiology. 2012;54(10):1109-18. doi: 10.1007/s00234-012-1023-y.

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With best regards,

Lukas Rasulić, Editor-in-Chief

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Dear colleagues and friends,

The Serbian Neurosurgical Society will mark the centennial anniversary of neurosurgery in Serbia with the main celebration and official ceremony on October 31st, 2023. During an eventful neurosurgery week from October 30th - November 3rd, 2023, we will hold the 9th SNSS Annual Meeting and the 10th SNSS Congress with international participation. We are delighted that we will have the opportunity to host a Joint ventures with several professional and continental associations, such as European Association of Neurosurgical Societies, World Academy of Neurological Surgery, Continental Association of African Neurological Societies, The Society of British Neurological Surgeons, Italian Society of Neurosurgery, International Academy of Neurosurgical Anatomy, and Southeast Europe Neurosurgical Society.

The global theme of the meeting, *"Focus. Dedication. Specialization",* sublimates the history of Serbian neurosurgery. The emergence of neurosurgery in Serbia officially dates back to 1923, when Milivoje Kostic, one of the pioneers in the field of surgery in this region, performed his first surgical treatment of a pituitary tumor via the nose. The way forward was further paved by his brother, Slobodan Kostic, who specialized in Stockholm in the early 1930s under Herbert Olivecrona, the leading European neurosurgeon at the time. Upon his return to Belgrade, Slobodan Kostic began applying all the basic principles of the Swedish Neurosurgical School, which was the highest standard in European neurosurgery.

On October 31st, 1938, Milivoje Kostic established a special Department of Neurosurgery within the Clinic of Surgery in Belgrade, and Slobodan Kostic was named the first head of the department. This was the first neurosurgery department in Serbia, and Yugoslavia, established only three years after the first neurosurgical department in Germany, in Wurzburg. Along with the centennial anniversary, we are also marking 85 years of the Clinic for Neurosurgery at the University Clinical Center of Serbia.

As we celebrate these important anniversaries, we share pride in our founders' achievements, and we use the opportunity to look back and to look forward. Serbian neurosurgery has long earned its place on the world map of neurosurgery. Leaning upon the strong foundations of tradition and experience and following contemporary professional accomplishments, trends and innovations, we strive for neurosurgery without frontiers and aim at building bridges of knowledge and understanding across disciplines, institutions, individuals, and generations.

Many things have changed in the first 100 years of neurosurgery in Serbia. But what remains a constant is our clear sense of purpose – continuous enhancement and international affirmation of neurosurgery in Serbia – always keeping in mind that the essence of our profession is, above all, the well-being of every individual patient.

We are honored that since the 1st SNSS Annual meeting in Belgrade in 2015, we have succeeded in increasing the visibility and presence of Serbian neurosurgeons in the international neurosurgical community and that many renowned international experts have accepted to attend our meetings.

The main topics to be explored in the 2023 SNSS meetings include neuro-oncology, neurovascular, neurotrauma & intensive care, spine & peripheral nerve, pediatrics, functional & stereotactic neurosurgery, and many more, aiming to highlight the state-of-the-art advances and challenges in the field.

Furthermore,

We are very glad that the relevant institutions of the Republic of Serbia have recognized the significance of neurosurgical education, and we appreciate their continued support.

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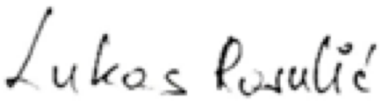
We will do our best to create a particularly memorable and inspiring intellectual and social experience in October 2023, and we look forward to welcoming you to Belgrade!

Cordially,



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